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Literacy Development in Children with Cochlear Implants

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Abstract

In general, school-age children with significant hearing loss demonstrate poorer literacy skills than their typically hearing (TH) peers. However, it is often difficult to infer from research findings which individual factors contribute to overall learning outcomes, given the high degree of heterogeneity in the hearing-impaired population. Since the development of any specialised intervention is based on evidence about the strengths and weaknesses of the targeted group, gaps in research arising from population variability have clinical and educational significance. Of the broader population of children who are deaf and hard of hearing, the research presented in this thesis pertains to a homogeneous group of beginning readers with significant bilateral hearing loss, who use cochlear implants (CIs) and communicate via solely spoken language. A psycholinguistic approach, founded on existing theoretical models, informed the collection and analysis of research findings in this thesis, which has thus allowed for specific interpretations as to the skills underlying reading and writing outcomes in children with CIs.

As described in Chapter 1, children with CIs have spoken and written language difficulties, relative to TH children. Chapters 2 and 3 of the present thesis expand on these findings, by comparing the behavioural performances of children with CIs with those of TH children, and by examining the underlying skills that contribute to observed literacy outcomes in each group. These results are complemented by research presented in Chapters 4 and 5, both of which use electroencephalography (EEG) to capture children's neural responses to simple linguistic tasks. In the thesis conclusion (Chapter 6), all study findings are integrated to provide a thorough, yet contextually informed, understanding of how literacy develops in children with CIs during the early school years.

The focus of Chapter 2 is on reading development in children with CIs who use spoken language. Results from a cohort of children with CIs were examined with reference to a TH control group, who were similar in age, gender and nonverbal reasoning ability. The CI group performed significantly worse than TH children on measures of reading accuracy and phonological processing. As expected, based on the Simple View of Reading model, word reading accuracy and listening comprehension contributed to reading comprehension for both groups. However, the predominant concurrent predictor of reading comprehension was word reading accuracy for the CI group and listening comprehension for the TH group. For all children, orthographic and phonological skills were found to contribute to word-level reading accuracy.

Chapter 3 examined the spelling skills of CI and TH children. With respect to overall accuracy, children with CIs achieved similar irregular and nonsense word spelling scores to the TH control group. Yet, spelling errors were less phonologically plausible for children with CIs, suggesting that they applied phonics knowledge less effectively when producing written words. In support of this interpretation, letter-sound knowledge was significantly related to nonword spelling performance for the TH group, but not the CI group.

In Chapter 4, EEG measures were used to examine semantic sensitivity to word-picture incongruence in the CI and TH groups. Comparisons were made on the basis that semantic processing skills – in combination with numerous other inter-related skills – contribute to overall word recognition and reading comprehension. Analysis of the results showed that a similar 'N400 effect', which is known to index sensitivity to lexical-semantic incongruence, was elicited in both the CI and TH groups. Hence, this fine-grained, semantically driven neural response was found to be normal in children with CIs.

Chapter 5 examined on-line rhyme sensitivity in TH children and a small group of children with CIs, again implementing EEG methodology to capture participants' neural responses. A letter rhyme judgement task elicited a significant 'rhyme effect' in children with typical hearing. Within this group, a larger amplitude of rhyme effect was significantly correlated with worse letter-sound knowledge, as measured behaviourally, and this relationship was mediated by nonverbal reasoning ability. Within the CI cohort, there was significant variability in the presence and size of rhyme effect elicited in each participant. At an individual level, those children who demonstrated a rhyme effect tended to have better nonverbal reasoning skills than those who did not demonstrate a rhyme effect.

Results from all four of the aforementioned studies contribute to a more complete understanding of how literacy develops in beginning readers with CIs. In the context of a classroom environment, the research findings suggest that children with CIs may show written language skill deficits, stemming in part from underlying phonological limitations.

Declaration by author

This thesis is composed of my original work, and contains no material previously published or written by another person except where due reference has been made in the text. I have clearly stated the contribution by others to jointly-authored works that I have included in my thesis.

I have clearly stated the contribution of others to my thesis as a whole, including statistical assistance, survey design, data analysis, significant technical procedures, professional editorial advice, financial support and any other original research work used or reported in my thesis. The content of my thesis is the result of work I have carried out since the commencement of my higher degree by research candidature and does not include a substantial part of work that has been submitted to qualify for the award of any other degree or diploma in any university or other tertiary institution. I have clearly stated which parts of my thesis, if any, have been submitted to qualify for another award.

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Publications included in this thesis

No publications included.

Submitted manuscripts included in this thesis

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Conference papers:

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Online news articles:

Bell, N. (2017, May 11). Explainer: How the brain changes when we learn to read. *The Conversation.* Retrieved from: https://theconversation.com/explainer-how-the-brain-changes-when-we-learn-to-read-76783.

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Contributions by others to the thesis

Anna Hyland helped to establish inter-rater reliability for the spelling error analysis in Chapter 3 by scoring approximately 50% of the total written productions.

Statement of parts of the thesis submitted to qualify for the award of another degree

No works submitted towards another degree have been included in this thesis.

Research involving human or animal subjects

The research presented in this thesis involved human subjects. No animal subjects were involved in this research.

Ethical clearance for the study was obtained from the Behavioural and Social Sciences Ethical Review Committee at the University of Queensland (2010000785). Gatekeeper ethical clearance was also obtained from the organisation Hear and Say.

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literacy, reading, spelling, phonology, cochlear implants, children, deaf, hearing loss

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1702 Cognitive Sciences (50%)

1109 Neurosciences (25%)

1103 Clinical Sciences (25%)

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List of Abbreviations

AVT	Auditory-verbal therapy
CC2	Castles & Coltheart 2
CELF-4	Clinical Evaluation of Language Fundamentals 4th ed.
CI	Cochlear implant
CMV	Cytomegalovirus
CNRep	Children's Test of Nonword Repetition
CTOPP-2	Comprehensive Test of Phonological Processing 2nd ed.
DiSTi	Diagnostic Spelling Test – Irregular Words
DiSTn	Diagnostic Spelling Test – Nonwords
DRC	Dual-route cascaded
EEG	Electroencephalography
EI	Early intervention
ERP	Event-related potential
HA	Hearing aid
LeST	Letter-Sound Test
LC	Listening comprehension
LR	Letter rhyme
LSK	Letter-sound knowledge
LVAS	Large Vestibular Aqueduct Syndrome
NS	Not specified
OP	Orthographic processing
PA	Phonological awareness
PDP	Parallel distributed processing
PM	Phonological memory
PPVT-IV	Peabody Picture Vocabulary Test 4th ed.
PTA	Pure tone average
RAN	Rapid automatised naming
RC	Reading comprehension
RE	Rhyme effect
RV	Receptive vocabulary
SD	Standard deviation
SLI	Specific Language Impairment

TH	Typically hearing
TOC	Test of Orthographic Choice
WR	Word reading
YARC	York Assessment of Reading Comprehension

Chapter 1. Literature Review:
Literacy Development in Children with Cochlear Implants

1.1. Introduction

Fundamental to literacy development in typically hearing children is phonological processing, or the ability to process sounds in spoken language (National Reading Panel, 2000; Wagner & Torgesen, 1987). This sensitivity allows for the formation of systematic correspondences between print and sound, which is a critical precursor to early word decoding (Catts, Herrera, Nielson & Bridges, 2015). Limitations in underlying phonological skills have been hypothesised to account for poor literacy outcomes, both in typically hearing children (Swan & Goswami, 1997), and in hearing-impaired children fitted with cochlear implants (Wang, Trezek, Luckner & Paul, 2008). This literature review will present evidence from existing research that has examined reading and spelling in individuals with cochlear implants. The theoretical models that guide our understanding of literacy development in typically hearing children will be described, and then applied to the population with cochlear implants, with consideration given to the individual and demographic factors that may have a mediating effect on outcomes. Finally, the potential for exploring aspects of literacy development through electrophysiological means will be explored, with specific regard to how this informs our current understanding of how literacy develops in children with cochlear implants.

1.2. Literacy Development in Children with Typical Hearing

There are a number of different theoretical models that may be used to represent the development of reading skills in typically hearing children. At the level of single word processing, the *dual-route cascaded* (DRC) model (Figure 1.1) describes three different routes for reading aloud (Coltheart, Rastle, Perry, Langdon & Ziegler, 2001). The lexical semantic and lexical nonsemantic routes (highlighted in Figure 1.1 in green and blue, respectively) generate pronunciation by way of accessing existing word representations. Using these routes, skilled readers may recognise familiar real words, regardless of the regularity with which letters in the word correspond with phonemes. Inclusion of the semantic system depends on whether information about word meaning is also retrieved. According to the DRC model, encountering unfamiliar or nonsense words necessitates the use of the sublexical route (highlighted in orange), which involves the application of known letter-sound correspondence rules. This is the route most used by early readers, for whom stored orthographic representations are likely to be less properly established (Leinenger, 2014).

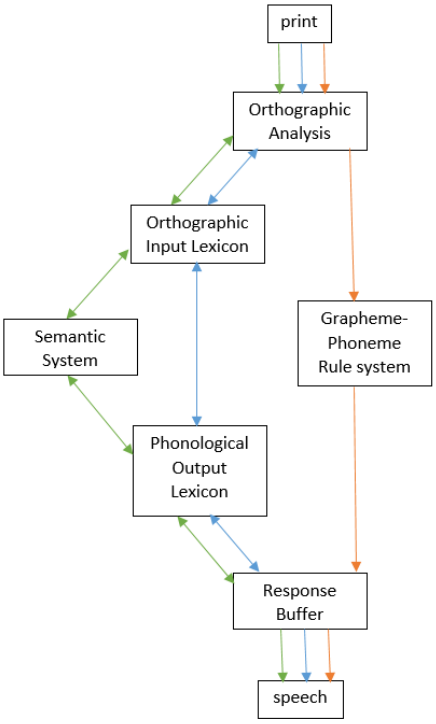

Figure 1.1. Components of the dual-route cascaded (DRC) model of single word processing.
Note. Green = lexical semantic route; blue = lexical nonsemantic route; orange = sublexical route. Adapted from Coltheart, Rastle, Perry, Langdon & Ziegler (2001).

As mentioned, the different routes presumed to exist in the DRC model are associated with different types of words. For skilled and unskilled readers alike, the pronunciation of a nonword such as 'norf' relies on the sublexical route, as this item has not been encountered before and, hence, does not exist as a stored lexical representation. A reader's ability to assemble pronunciations based on knowledge of letter-sound correspondences may be assessed by measuring their nonword reading accuracy (Coltheart et al., 2001). On the other hand, their ability to retrieve and pronounce whole lexical representations may be assessed by measuring their irregular word reading accuracy on items such as 'yacht'. These words 'disobey' letter-sound correspondence rules, and as such cannot be pronounced via the sublexical route (Coltheart et al., 2001).

A competing, connectionist approach describing the processes underlying word recognition is theorised in the *parallel distributed processing* (PDP) model (Figure 1.2). The framework, as outlined by Seidenberg and McClelland (1989) assumes that successful single word reading relies on the simultaneous computation of orthographic, semantic and phonological codes. The interactivity inherent in this approach means that each type of activated representation influences – and is influenced by – other representations. This aspect of the model contrasts with the DRC model, in which the activation of phonological coding is not thought necessary for word retrieval, except via the sublexical route, which is involved in decoding unfamiliar items (Leinenger, 2014). A set of 'hidden units', represented in Figure 1.2 as blank circles, reflects the mediatory connection between areas (Seidenberg, 2005). In the context of reading development, the model assumes that the networks underlying efficient word recognition become increasingly refined through continued exposure to phonological, orthographic and semantic representations (Harm & Seidenberg, 2004; Seidenberg, 2005).

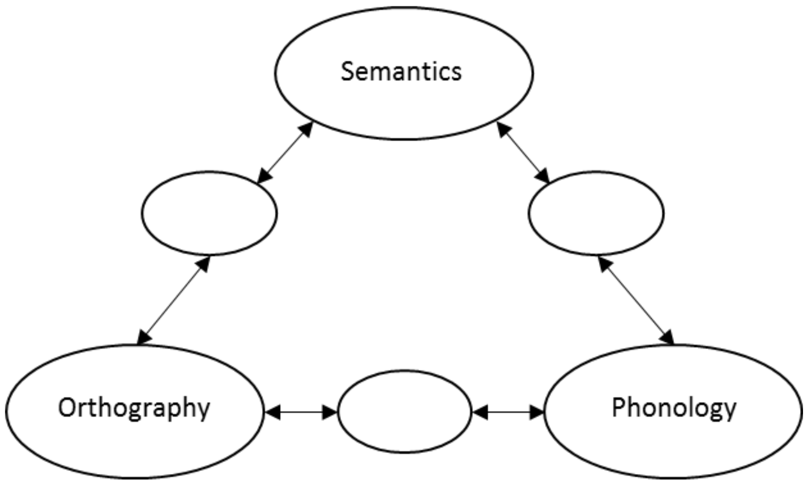

Figure 1.2. Components of the parallel distributed processing (PDP) model of single word processing.
Note. Adapted from Seidenberg (2005).

In contrast with the dual-route approach, the PDP model does not assume that separate routes underlie the recognition of real and nonsense words. However, areas of the model may be differentially weakened in cases of acquired and developmental dyslexia, resulting in similar patterns of performance as those described by the authors of the DRC model (Manis, Seidenberg, Doi, McBride-Chang & Peterson, 1996). Deficits in nonword reading may be attributed to impaired phonological representations, while reduced irregular word reading accuracy may be attributed to an impaired connection between orthographic and phonological representations (Manis et al., 1996). By this account, the same processes of excitation and inhibition underlie the processing of all types of letter-strings, regardless of regularity and lexicality. Yet, the degree to which each area of knowledge contributes to overall word retrieval is separable, potentially observed as different patterns of performance across word types.

Skills associated with word recognition and listening comprehension are captured by the *Simple View of Reading* model, which provides a general framework for reading comprehension development (Catts, Adlof & Weismer, 2006; Hoover & Gough, 1990). It is assumed within the model that in order to comprehend the content of a written text, the reader must be able to decode or recognise words, and must also have the linguistic knowledge required for attaching meaning to the words, within their context. According to the Language and Reading Consortium (2015), while the processes that contribute to reading comprehension are numerous, varied and interactive, their effects are largely indirect, acting predominantly via the broader skill areas of word recognition and listening comprehension.

One important aspect of the Simple View of Reading model is that relative weightings of the reading comprehension sub-skills are age-dependent. The role of word recognition ability is most significant in early readers, for whom attentional resources and literacy instruction are largely devoted to decoding single words (Catts, Hogan & Adlof, 2005; Catts et al., 2015; Language & Reading Consortium, 2015). More specifically, Catts and colleagues (2015) recently reported that kindergarten-level letter knowledge and phonological awareness were indirectly predictive of third-grade reading comprehension, through their relationships with second-grade word reading accuracy. Over time, the demands associated with successful reading comprehension are seen to shift, so that higher level vocabulary and spoken language skills are most predictive of reading comprehension in Grade 2 and beyond (Catts et al., 2005). This is presumably due to the age- and experience-dependent development of word recognition automaticity (Language & Reading Consortium, 2015). When determining the predictive value of reading comprehension sub-skills, it is crucial to account for these developmental changes, in order to guide expectations of what constitutes ‘normal’ reading.

Both of the aforementioned DRC and PDP models of single word recognition necessitate a role for phonological processing. The influence of this factor in typical early reading development has been well established over the course of the last few decades (see Castles & Coltheart, 2004, for review). Abundant research has shown that the ability to identify and manipulate individual word segments, referred to as ‘phonological awareness’, is predictive of later reading outcomes (Badian, 1998; Hulme et al., 2002; Wagner, Torgesen & Rashotte, 1994). Similarly, the explicit targeting of phonological awareness has been seen to enhance reading outcomes over time when it is combined with instruction on letter-sound correspondences (Ball, 1997; Bradley & Bryant, 1983).

The same theoretical underpinnings of reading development are sometimes also applied to the study of spelling. Accordingly, the partial separability of underlying phonological, orthographic and semantic sub-skills means that, as with reading, patterns of inaccuracy in nonword and irregular word spelling have been associated with weaknesses in sublexical and lexical processing mechanisms, respectively (Brunsdon, Coltheart & Nickels, 2005; Cholewa, Mantey, Heber & Hollweg, 2008). Thus, for both reading and spelling, it may be valuable to assess an individual's accuracy across various word types, since different profiles may emerge which indicate the presence of specific weaknesses in underlying literacy sub-skills. In particular, and in accordance with proponents of both cascading (e.g., Figure 1.1) and interactive (e.g., Figure 1.2) theoretical models, nonword spelling (and reading) difficulties may be attributed to weak phonological processing abilities in typically hearing children (Bullinaria, 1997; Cholewa et al., 2008).

1.3. Literacy Development in Children with Hearing Loss

Children with hearing loss have often been reported to lag behind their typically hearing peers, in terms of literacy skill development (see Harris, 2015, for review). A study by Nelson and Crumpton (2015) examined the literacy skills of children with varying degrees of hearing loss, using a comprehensive battery of sound-, word-, sentence- and discourse-level assessment measures. They found that the participants, aged 6 to 18 years old, performed significantly worse than their typically developing individually-matched peers on all measures, apart from written expression at the discourse level. Areas of deficit included reading comprehension, as well as single word reading accuracy and spoken language comprehension. Importantly, the children with hearing loss were relatively heterogeneous, in terms of age at assessment, age at hearing loss identification, and age at access to hearing technology. Still, the findings reported by Nelson and Crumpton (2015) clearly highlight a disparity in literacy achievement between hearing-impaired and typically hearing paediatric populations.

Advances in cochlear implant (CI) technology have enabled children with severe-to-profound hearing loss to access spoken language. There is a large body of evidence showing that the language-learning trajectories of children with significant hearing loss improve after receiving the implant (e.g., Leigh, Dettman & Dowell, 2016; Tait, Nikolopoulos & Lutman, 2007). However, it is difficult to compare these outcomes with those for children who use other hearing technologies. Cochlear implantation surgery in some parts of the world is available only to individuals whose severity of hearing loss is so significant that they do not respond sufficiently to hearing aids (Houston, Vilga, Bradham, Teagle & Cunningham, 2018). Hence, the severity of participants' hearing loss will likely confound results in any comparative study where it is not kept equivalent across groups, thereby potentially concealing the positive effects of CI technology.

Where unaided hearing thresholds signify profound degrees of hearing loss across groups of children with different hearing devices, CIs are associated with better outcomes than hearing aids, on measures of receptive and expressive language (Tomblin, Spencer, Flock, Tyler & Gantz, 1999) and speech production (Horga & Liker, 2006). Given the importance of spoken language to typical literacy development, it may be expected that such results will have beneficial flow-on effects to reading and spelling. On the other hand, Harris and Terlektsi (2011) reported significantly better reading comprehension outcomes in older school-aged children with hearing aids, compared with children with CIs who were the same age and had the same degree of severe-to-profound hearing loss. The authors attributed this finding to the educational environment, which was another factor differentiating the groups. This conclusion serves as a valuable reminder of the hearing-impaired population's heterogeneity, which can be problematic when attempting to attribute outcomes to just one specific factor. Further research into the levels of literacy success obtainable through specific hearing technologies is therefore required, especially as device features continue to improve over time.

1.3.1. Literacy development in children with cochlear implants. Since the onset and continued advancement of CI technology, many studies have examined literacy skills in this specific subset of the hearing-impaired child population. Candidacy for cochlear implantation is evaluated on an individual basis, but CI users typically have significant (i.e., severe-to-profound) sensorineural hearing loss (Cochlear, 2018; Zwolan, 2015). As with the rest of the deaf and hard-of-hearing population, children who have received CIs often demonstrate impaired reading comprehension, compared with their typically hearing peers. This finding has been reported for emergent readers (Nittrouer, Caldwell, Lowenstein, Tarr & Holloman, 2012), older school-aged children (Geers, 2003; Nittrouer, Caldwell-Tarr, Sansom, Twersky & Lowenstein, 2014; Weisi et al., 2013) and adolescents (Geers & Hayes, 2010; Harris & Terlektsi, 2011). Nittrouer and colleagues (2012) also reported widespread difficulties for the CI group on a number of phonological and reading measures. The authors attributed the observed reading difficulties to decreased phonological awareness, resulting from acoustic processing limitations in the cochlear device itself.

Indeed, there is a significant amount of evidence showing that children with CIs experience difficulty with performing phonological awareness tasks (Ambrose, Fey & Eisenberg, 2012; James, Rajput, Brinton & Goswami, 2008; Lee, Yim & Sim, 2012; Nittrouer et al., 2012; 2014; Spencer & Tomblin, 2009; Weisi et al., 2013). Ambrose et al. (2012) measured the literacy precursor skills of preschool-aged children with CIs, in comparison to typically hearing children of the same age. Phonological awareness was measured using phoneme elision (e.g., ‘point to “team” without /m/’) and blending tasks (e.g., ‘what word do these sounds make: /f/ – “ox”?’). The researchers found that the phonological awareness skills of children with CIs were more than one standard deviation below the mean of their peers. In contrast with these findings, there were no significant differences between groups on measures of print knowledge. Nittrouer and colleagues (2014) demonstrated similar deficits in phonological awareness performance in older school-aged readers with CIs. Their sample of 8-year-old children performed significantly worse on matching and elision tasks, which relied on awareness of the target’s phonemic structure.

In the same study by Nittrouer and colleagues (2014), the greatest discrepancy in performance between CI and typically hearing groups was demonstrated on a nonword repetition task. Accurate nonword repetition requires access to phonological representations, without reference to metalinguistic knowledge or information pertaining to existing lexical items (Stone & Brady, 1995). A number of other studies have also reported significant nonword repetition difficulties in children with CIs (Geers & Hayes, 2010; Lee et al., 2012; Spencer & Tomblin, 2009). Nittrouer and colleagues (2014) concluded that poor nonword repetition was representative of the CI group’s relatively undefined phonological representations. In support of this theory, populations with dyslexia generally demonstrate similar difficulties with nonword repetition (Melby-Lervåg & Lervåg, 2012), as well as phonological awareness (Swan & Goswami, 1997).

Given the observed difficulties in sublexical decoding and phonological processing, it may be expected that word reading accuracy will also be reduced in children with CIs. Such a prediction is based on the self-teaching hypothesis, which assumes that lexical representations in typically hearing children develop as a partial consequence of phonological recoding, or the repeated application of phoneme-grapheme correspondences (Share, 1995).

Indeed, children with CIs assessed on word reading accuracy and phonological processing measures have often shown reduced performances in both assessment tasks (Harris & Terlektsi, 2011; Johnson & Goswami, 2010; Nittrouer et al., 2014; Weisi et al., 2013). However, in a potential exception to this pattern, Fitzpatrick and colleagues (2012) reported that children with CIs achieved an overall word reading standard score of 98.7, while phonological awareness was 75.9 and phonological memory as low as 66.9. These findings would appear to indicate that reading accuracy can develop on the basis of 'sight word' learning and orthographic processing, as opposed to phonological processing. Yet, the authors also stated that 'more than half' of their CI group obtained age-appropriate scores in the word reading measure, which suggests that, despite the relatively normal mean score, a significant proportion of the group performed below normal limits. Debate regarding the role of phonology in predicting reading outcomes is ongoing, as will be discussed in Section 1.4 of this literature review.

When evaluating the literacy abilities of children with CIs, it is important to consider spelling performance, as well as reading. Spelling measures have been less frequently included in prior studies involving children with CIs. Yet, given that accurate spelling often relies on sensitivity to the word's phonological features, single word spelling assessment may serve to provide valuable information about an individual's literacy skills (Hayes, Kessler & Treiman, 2011). In general, children with CIs show small but significant deficits in spelling achievement. Apel and Masterson (2015) analysed the spellings of nine children with CIs, who had a mean age of 8;11 years. Accuracy for this group was poorer than that observed in a typically hearing group of children matched on reading ability, although the difference between cohorts did not reach statistical significance. Standardised spelling scores were not reported by the authors, which limits the validity with which spelling accuracy for children with CIs could be compared with age-based expectations. In addition, the CI group comprised a combination of sign, spoken and total language users (Apel & Masterson, 2015). Since communication mode has previously been found to influence literacy outcomes (see Section 1.5.2; Cupples et al., 2017; Johnson & Goswami, 2010; O'Donoghue, Nikopoulos & Archbold, 2000), the skills of spoken language users may be better represented when analysed in isolation.

Roy, Shergold, Kyle and Herman (2015) investigated the spelling abilities of 10- and 11-year-old spoken language users with severe-to-profound hearing loss, with comparative reference to a dyslexic group, as well as the age-based norms for the test. The authors combined children with hearing aids and CIs, based on the finding that there was no significant difference between subgroups. They reported that performances on both reading and spelling measures were significantly below age expectations for the hearing-impaired group, although scores were, on average, within one standard deviation of the normative mean. In a similar study, Hayes, Kessler and Treiman (2011) compared the spelling results of a 6- to 12-year-old CI cohort with their individually age-matched hearing peers. Despite finding 55% spelling accuracy in CI children and 66% accuracy in typically hearing children, the difference between groups was not significant. However, accuracy results were only analysed once reading comprehension had been statistically controlled. It is possible that this step of controlling for reading ability obscured group differences, since reading and spelling are intrinsically linked and share fundamental underlying processes (Puranik, Lonigan & Kim, 2011). More research into spelling development would help to establish whether children with CIs, who use solely spoken communication, show difficulties in this area of literacy, when compared with their age-matched peers.

1.4. Relationships Between Literacy Sub-skills in Children with Hearing Loss

Within the broader population of individuals with hearing loss, some research has been conducted to look at the relationships between skills involved in literacy development. Mayberry, del Giudice and Lieberman (2011) conducted a meta-analysis to investigate the role of phonology in the reading development of individuals with severe-to-profound hearing loss. Participants included for analysis ranged from 4 to 62 years of age. The authors found that 'phonological coding and awareness' scores predicted only 11% of the variance in reading performance, while 'language ability' predicted 35% of the variance. These findings were interpreted as meaning that phonological skills are not critical in attaining successful reading outcomes. Such an argument has been posited by a number of researchers, who claim that literacy skills develop differently for children with significant hearing loss (Allen, Clark, del Giudice, Koo & Lieberman, 2009; Mayberry et al., 2011; Miller & Clark, 2011). This theory is based on the premise that phonological awareness may essentially be 'bypassed', and that lexical representations may be formed and retrieved based on existing linguistic knowledge and holistic orthographic information.

There are, however, some key limitations of the Mayberry et al. (2011) meta-analysis that are worth noting. Firstly, the actual proficiency of reading skill was often not accounted for in the studies, which throws doubt onto the claim that high-level (or even average) reading achievement can be attained in the absence of adequate underlying phonological ability. In addition, the studies reviewed in the meta-analysis included a wide range of ages, which does not allow for the change in reading profile that may occur over time. That is, for beginning readers, the Simple View of Reading model holds that word recognition and phonological processing are more predictive of reading comprehension ability than listening comprehension (Catts et al., 2006; Garcia & Cain, 2014; Language & Reading Consortium, 2015). With time, word recognition becomes more automatic, and the role of spoken language is seen to gain prominence (Language & Reading Consortium, 2015). The issue of sample age is therefore a critical factor that should be considered when analysing the relative statistical weightings of reading sub-skills to predict overall reading comprehension. Due to the heterogeneity inherent in the hearing-impaired population, it is not surprising that many studies have conducted regression-based analyses into reading comprehension, using a sample of children who fall within a wide age range (e.g., Johnson & Goswami, 2010; Nelson & Crumpton, 2015; von Muenster & Baker, 2014). Such findings, however, need to be interpreted with caution.

In contrast to the claims made by Mayberry et al. (2011), many researchers have suggested that the role of phonology is just as crucial for children with hearing loss as it is for children with typical hearing (Mayer & Trezek, 2014; Wang et al., 2008). The concurrent relationship between reading comprehension and phonological processing in early readers with varying degrees of hearing loss was examined by Cupples, Ching, Crowe, Day and Seeto (2013). Although their sample of aided pre-literate children showed relatively reduced performances on measures of phonological awareness, scores on this task were still predictive of early reading skills, after controlling for nonverbal IQ, receptive language, and a number of demographic variables. Similarly, Webb, Lederberg, Branum-Martin and Connor (2015) found that word reading performance in early readers with hearing loss (ranging from mild to profound) was best explained using a 3-factor model, in which phonological awareness, letter knowledge, and vocabulary skill were all included as separate but highly correlated constructs.

In typically hearing children, longitudinal research provides strong evidence in support of the theory that phonological processing is a causal factor in facilitating successful word reading development (Hulme et al., 2002; Wagner et al., 1994). The longitudinal relationship between phonological processing and word recognition in children with severe-to-profound hearing loss was investigated in an early study by Harris and Beech (1998). At the initial assessment, when participants were 5 years old, they were administered an implicit phonological awareness task in which they selected two pictures from a choice of three that had matching initial, medial or final sounds. Scores from this task were significantly correlated with growth in word reading comprehension, measured after one year of reading instruction ($r = 0.43$; $p < 0.05$). In another study with similar aims, Colin, Magnan, Ecalle and Leybaert (2007) found that implicit phonological processing performance measured in pre-literate children significantly predicted Grade 1 word recognition, in those with severe-to-profound hearing loss ($r^2 = 0.283$; $p = 0.01$) and in those with typical hearing ($r^2 = 0.311$; $p = 0.01$). Together, these studies suggest that phonological processes significantly contribute to literacy abilities in children with significant hearing loss.

1.4.1. Relationships between literacy sub-skills in children with cochlear implants. With specific regard to the population of children with CIs, the aforementioned pattern of results is complemented and expanded upon by Nittrouer and colleagues (2012). They found that phonological awareness concurrently predicted 54% of the variance in word reading scores for preschool children with CIs and 68% of word reading variance for typically hearing children. Hence, phonological processing did appear to be a significant precursor to word reading abilities, regardless of hearing status. At the same time, overall reading comprehension in the CI group was predicted more by measures of auditory comprehension and expressive vocabulary, while for the control group it was predicted by phonological awareness scores and vocabulary. Given that reading outcomes were generally better in the control group, it may be posited that early reading comprehension for children with CIs was carried out with comparatively less success, and with comparatively less input from phonological processing mechanisms.

The role of phonological processing in reading has also been examined in older samples of children, with results suggesting that deviant reading profiles may be associated with poorer outcomes in those with CIs. James, Rajput, Brinton and Goswami (2009) found that receptive vocabulary accounted for more unique variance in word reading accuracy in 8-year-old children with CIs (45%), compared with age-matched typically hearing children (18%). For both groups, rhyme awareness also significantly predicted word reading (21% and 28% for children with CIs and their hearing peers, respectively). Interestingly, in a group of 6- and 7-year-old typically hearing children matched with the CI group on reading skill level, rhyme awareness was the only significant predictor of word reading accuracy (45%). Hence, the younger, typically hearing group appeared to be largely reliant on phonological processing mechanisms to achieve the same reading accuracy as that obtained by children with CIs, who were seemingly more reliant on vocabulary. As with the study by Nittrouer and colleagues (2012), these findings point towards qualitative differences in the reading profiles of hearing-impaired children with CIs. Notably, the children with CIs in James et al.'s (2009) study were implanted comparatively late, at a mean age of 4;7 years. Given that literacy outcomes are influenced by age at implantation (discussed in Section 1.5.1), different results may potentially be expected with an earlier-implanted sample of children.

In another study involving a similarly aged cohort, children with CIs aged between 8 and 12 years performed a lexical decision task, wherein they judged whether target stimuli were real words (Bouton, Colé, Serniclaes, Duncan & Giraud, 2015). Although the CI group rejected pseudohomophones (i.e., nonwords that sound like real words, e.g., 'faik') with the same accuracy as typically hearing reading- and age-matched controls, they were significantly less accurate with rejecting non-homophonic nonwords (e.g., 'kemp'). The authors suggested that the retrieval of phonological representations was less automatic for children with CIs, resulting in an over-reliance on whole-word orthographic processing (Bouton et al., 2015). Difficulties at this level may be attributed to underlying limitations in phonemic perception, based on evidence from the same study showing group differences on a minimal pair discrimination task.

With regard to spelling performance, children with CIs are generally found to produce a greater proportion of spelling errors that are phonologically 'implausible' (i.e., misspellings that, if read aloud, do not reflect the target item's phonetic pronunciation). In the previously mentioned study by Roy et al. (2015), the proportion of phonologically implausible spelling errors produced by the combined group of hearing aid and CI users was significantly higher than in groups of dyslexic and typically developing children. Similarly, Hayes and colleagues (2011) found that children with CIs who used spoken language produced more phonologically implausible errors than typically hearing children. This pattern of results suggests that the population of children with CIs may not always utilise a phonological route for spelling. However, given the observed deficits in literacy performance, this does not necessarily provide support for the argument that successful literacy outcomes can be attained without adequate phonological processing abilities. Based on the findings from this study and others, it may be posited that some older children with CIs have developed an adequate degree of word recognition knowledge, using linguistic context and awareness of holistic lexical information to compensate for phonological processing deficiencies, although on average, these skills appear to be below the standards of their typically hearing peers.

Evidence for the important role of phonological processing in reading and spelling task performance should not be seen to undermine the significant predictive value of oral linguistic skill. As described previously, successful comprehension of a written text requires a number of inter-related and experience-dependent processes, including both word recognition and receptive language (Language & Reading Consortium, 2015). With regard to the latter set of abilities, a number of studies involving children with CIs have reported reduced performances on measures of receptive language (Ceh, Bervinchak & Francis, 2013; Ching, Day & Cupples, 2014; Fitzpatrick et al., 2012; Spencer, Barker & Tomblin, 2003). For example, 9- and 10-year-old children assessed by Spencer and colleagues (2003) demonstrated significantly reduced performances on both receptive and expressive language measures, when compared with typically hearing age-matched peers. The cohort were a mix of oral and sign communicators, and they were implanted comparatively late in life, at a mean age of 3.9 years.

Ceh and colleagues (2013) reported similar spoken language difficulties in emergent readers with CIs, all of whom received their implants before turning 2 years old. On a composite measure of receptive language, the group demonstrated an average delay of 11.7 months (SD = 14.3), compared with the standardised test norms. Interestingly, the same group performed within normal limits in early reading ability, although it should be noted that this was based on a composite measure of alphabetic knowledge, print convention knowledge and reading comprehension. Given that children with CIs have demonstrated comparatively normal abilities in alphabetic and print convention composite skill measures (Ambrose et al., 2012), it is possible that skills in reading were over-estimated. Alternatively, early access to sound via a CI could have resulted in more positive long-term reading outcomes. Nevertheless, it would appear that children with CIs show spoken language difficulties, which, like phonological processing difficulties, may be predicted to have flow-on effects to literacy achievement, according to the Simple View of Reading model.

Within the Simple View of Reading model, it is assumed that receptive language scores represent the ability to apply lexical knowledge to sentence- and discourse-level contexts (Language & Reading Consortium, 2015). The semantic quality of the lexical representation itself is therefore a critical supporting factor, which allows for the successful comprehension of stimuli. A study published by the Language and Reading Consortium (2015) showed that in typically hearing school-aged children, vocabulary was indirectly predictive of reading comprehension, via listening comprehension skill and, to a lesser extent, via word recognition skill. In children with CIs, a number of studies have shown that vocabulary scores are below the standards of their typically hearing age-matched peers (Ching et al., 2014; Geers & Hayes, 2010; James et al., 2008; Johnson & Goswami, 2010; Nittrouer et al., 2012; 2014). Again, these deficits may therefore influence outcomes in reading and spelling. Moving beyond the relationships between different literacy skills, there are also demographic and individual factors that impact on reading development in children with hearing loss.

1.5. Factors Influencing Literacy Success in Children with Hearing Loss

1.5.1 Age at cochlear implantation. The age at which individuals with hearing loss receive CIs has often been found to influence later literacy success (Archbold et al., 2008; Connor & Zwolan, 2004; James et al., 2008; Johnson & Goswami, 2010; Weisi et al., 2013). This relationship is attributed in some degree to the limited quality and quantity of auditory exposure received in very early infancy if children are not optimally amplified (Ruben, 1997). The first year of post-natal life is especially critical for the development of phonemic sensitivity and discrimination skills, which form the foundation for spoken language growth (Ruben, 1997).

There exists a large body of literature to suggest that early speech and language development is influenced by age at cochlear implantation (Colletti, Mandalá, Zoccante, Shannon & Colletti, 2011; Cupples et al., 2017; Dettman et al., 2016; Leigh, Dettman & Briggs, 2013; Tait et al., 2007; Tomblin, Baker, Spencer, Zhang & Gantz, 2005). Dettman and colleagues (2016) conducted an extensive study, involving over 400 children with CIs. Age at implantation for their sample ranged from under 12 months to 6 years, and multiple regression analyses revealed that this factor significantly predicted performances on school-age measures of speech perception, speech production, receptive vocabulary, and spoken language. Furthermore, a greater percentage of children implanted before 12 months obtained ‘normal’ standard scores on the same measures of speech production, receptive vocabulary and spoken language, thus providing evidence in support of early cochlear implantation. It may, however, be observed that the study did not account for differences in participants’ linguistic modalities. According to Harris (2015), children implanted earlier more commonly use spoken language to communicate, while those implanted later make greater use of sign language and speech-reading. Hence, it is not clear whether the positive outcomes observed for early implantees in the study by Dettman and colleagues (2016) are influenced by the group’s choice of communication modality.

A number of studies have found that, like speech and language outcomes, the literacy outcomes of school-aged children with CIs are affected by age at implantation (Archbold et al., 2008; James et al., 2008; Johnson & Goswami, 2010; Weisi et al., 2013). James and colleagues (2008) compared ‘early’ (implanted) and ‘late’ (implanted) 7- to 9-year-old children with CIs against each other, and against reading- and age-matched peers. They found that the ‘early’ group (implanted at a mean age of 2.1 years) outperformed the ‘late’ group (implanted at a mean age of 6.0 years) on measures of phonological awareness, receptive vocabulary and word reading accuracy. Similarly, Weisi and colleagues (2013) included age at implantation as a potentially predictive factor in multiple regression analyses, and found that it contributed significant variance to composite reading ability scores in 7- and 8-year-old readers implanted at or before the age of 3 years old.

Contrary to this pattern of results, Dunn and colleagues (2014) longitudinally studied the role of age at implantation in reading, and found that its predictive effect on reading comprehension just failed to reach significance at age 7 ($p = 0.054$). In this study, ‘early’ implantees received cochlear devices before they were 2 years old, while ‘late’ implantees received them between the ages of 2 and 4 years. Other studies have also failed to find a significant relationship between word reading ability and age at implantation (Cupples et al., 2013; Geers, 2003).

Clearly, the evidence with regard to the true effects of age at implantation is mixed, although there does appear to be a general trend for better outcomes in children implanted early. Continued research is required, in order to clarify the role of this and other interacting variables. In addition, it should be noted that, with the advancement of technology and the increase in support for early implantation, definitions of what constitutes ‘early’ and ‘late’ implantation have been seen to change over time. In Australia, it is now common for children with severe-to-profound hearing loss as young as 6 months old to undergo cochlear implantation surgery (Dettman et al., 2016). Still, research into the literacy outcomes of children implanted with cochlear devices before the age of 18 months is limited, and future research is warranted to examine how this cohort develops.

1.5.2. Communication mode. Even with the benefits of early implantation, there are additional factors influencing the outcomes of children with CIs. Another significant determinant of later success is the linguistic environment in which the child develops, during the early years after implantation. The increasing effectiveness of hearing aid and CI technology has resulted in a shift in how children with significant hearing loss communicate, such that there is now the option for them to access spoken language, instead of – or in combination with – sign language. Some authors have reported that reading outcomes were significantly influenced by the primary mode of communication used by participants with CIs (Johnson & Goswami, 2010; O'Donoghue et al., 2000). Other studies, however, have not shown a significant difference in the performances of oral and 'total' communicators, the latter category including those children who use a combination of spoken and sign language (Connor & Zwolan, 2004; Geers, 2003). Recently, a systematic review was conducted to examine the speech and language outcomes of children with varying degrees of hearing loss, who used different communicative modes (Fitzpatrick et al., 2016). The authors concluded that there was insufficient evidence to determine whether a combination of spoken and sign language was any more beneficial than spoken language in isolation.

In an effort to provide such evidence, and thus to directly address whether communicative mode affects developmental outcomes, Geers, Mitchell, Warner-Czyz, Wang and Eisenberg (2017) conducted a large-scale study involving children with CIs. The children were grouped according to whether they had no history of using sign language, a short term-history of using sign language (i.e., stopped within two years of receiving CIs), or a long-term history of using sign language (i.e., continued at least three years after receiving CIs). There were no significant differences between these groups with respect to gender, family income, age at aiding, average hearing threshold, age at cochlear implantation, or nonverbal cognition. Between 5 and 8 years old, children without sign language exposure showed better spoken language skills than those with a long-term history of sign language use. Between 9 and 12 years old, children without sign language exposure showed better spoken language skills than both sign language groups, and better reading scores than the long-term sign language group. Hence, spoken communication in isolation was related to better developmental outcomes, and indeed the skills of this group appeared to improve with age, relative to the skills of children with a history of sign language use. Importantly, it may be noted that analyses into the effects of communication mode provide only some insight into the form of language instruction to which children with significant hearing loss are actually exposed. In other words, when evaluating the influence of linguistic environment, it is worth investigating the aims and outcomes of specific behavioural interventions.

1.5.3. Exposure to behavioural intervention. One intervention option with an emphasis on promoting spoken communication is auditory-verbal therapy (AVT). Key to this program is the provision of auditory-verbal linguistic stimuli, which is not supplemented with any sort of sign or visual speech cue (White & Brennan-Jones, 2014). A series of studies by Dornan and colleagues tracked the longitudinal outcomes of children with moderate-to-profound hearing loss who underwent AVT (Dornan, Hickson, Murdoch & Houston, 2007; 2009; Dornan, Hickson, Murdoch, Houston & Constantinescu, 2010). Reading comprehension outcomes, as reported in their 2010 paper, showed that the hearing-impaired AVT sample scored within the 83rd percentile at approximately 7 years of age, and within the 88th percentile a year later. These outcomes were comparable to a typically hearing group of children, who were matched on gender, socio-economic status, receptive vocabulary and spoken language scores, but not chronological age. It should be noted there were only seven participants in each group that had entered formal schooling. Hence, due to the small sample size, no statistical comparisons could be made between groups. Further research investigating the outcomes of AVT would therefore be valuable.

The authors of the above studies defended their use of a language-matched control group, stating that the aim was to track the progress of children with hearing loss and compare this growth with a typical developmental trajectory. If groups had been matched on age, the typically hearing children would have had a higher language baseline score, which may have distorted results (Dornan et al., 2010). While the published findings are indeed important, there exists the opportunity for future research to explore the speech, language and literacy skills of children who have received AVT intervention, compared with those obtained from an age-matched control group. This would provide insight into their abilities within the context of developmental expectations.

Another key principle of AVT is the alignment of developmental goals with the typical progression and patterns observed in typically hearing children (White & Brennan-Jones, 2014). Implicit in this perspective is the notion that literacy learning develops along the same trajectory for all children, regardless of hearing status. In other words, the phonological processes long theorised to underlie early literacy knowledge in typically hearing children should be just as integral to children with a hearing impairment. This same principle is emphasised in intervention strategies which aim to enhance phonological awareness through visual means. For example, speech-reading has been found to significantly relate to literacy development in children with severe or profound hearing loss (Harris & Moreno, 2006; Johnson & Goswami, 2010; Kyle & Harris, 2006). According to Johnson and Goswami (2010), some children may be able to obtain phonological information from a visual representation of the target phoneme’s articulatory placement. In this way, Harris

(2015) suggests that phonological skills may be best thought of as being multi-modal, instead of exclusively aural (see also Leybaert & LaSasso, 2010).

Cued Speech and Visual Phonics programs explicitly teach letter-sound correspondences through the use of specific hand and mouth movements (Wang et al., 2008). A study by Trezek and Malmgren (2005) looked at the pre- and post-treatment outcomes of children with varying degrees of hearing loss, who were randomly allocated to either a Visual Phonics-focused intervention or a comparison group. Following twenty sessions targeting phonics and phonemic awareness skills, the treatment group performed significantly better on measures of phonological awareness and nonword reading measures. Follow-up studies by Trezek and colleagues (Trezek & Wang, 2006; Trezek, Wang, Woods, Gampp & Paul, 2007) showed significant improvement in the treatment group's word reading, reading comprehension, and spelling performances, as measured using standardised assessments. These findings support the theory that children with and without hearing loss follow the same developmental trajectory, and that the explicit targeting of phonics and phonological awareness skills results in similarly enhanced literacy performances.

Given the number of demographic and audiological factors that may vary among children with hearing loss, it is perhaps unsurprising that there is limited research into intervention outcomes of children with CIs. That said, one study by von Muenster and Baker (2014) analysed the relationships between reading sub-skills in children with CIs who were exposed to AVT. According to their findings, phonological awareness skills were significantly and strongly correlated with measures of reading comprehension and accuracy, indicating that those children who demonstrated better phonological processing abilities had better literacy outcomes. As with most of the other research reviewed in this paper, these results support the theory that phonological processing plays a critical role in successful reading development.

Unfortunately, the study by von Muenster and Baker (2014) did not compare the group's scores with standardised norms or a typically hearing control group, so it is unclear how the combined effects of AVT and cochlear implantation would have been reflected in the context of age-based expectations. However, given the success with which ongoing advances in CI technology enable increased phonetic discrimination, it may be hypothesised that AVT will complement the effects of cochlear implantation, by exposing children to targeted, phonologically-based aural instruction (Dornan et al., 2010; although see Leybaert & LaSasso, 2010, for audiovisual perspective on phonological development).

1.6. Electrophysiological Measures of Language and Literacy

1.6.1. Electroencephalography and event-related potentials. All of the studies previously described in this review have obtained evidence using behavioural task measures. This method of investigation is both practical and clinically valuable, as the information gathered may be applied to observable behaviours in the participant's or population group's real life. Patterns of performance may also be interpreted within the context of current understanding about children's developing neural circuitry. Ergo, based on the observation of a comparatively reduced performance in one assessment task, it may be assumed that the processes thought to underlie that performance are under-developed or deviating in some way from the norm. Such assumptions are complemented by evidence from direct measures of neurological function. Through methods such as electroencephalography (EEG), on-line and real-time neurological data can be recorded, thereby enabling more accurate and specific links to be established between surface-level behavioural responses and underlying cognitive processes.

Electroencephalography is sometimes used in combination with techniques to elicit event-related potentials (ERPs), which are indices of significant changes in neurological activity resulting from exposure to a time-locked stimulus (Kutas, Van Petten & Kluender, 2003). Different ERP 'components', elicited by specific task conditions, may be observed on EEG waveforms and are characterised by the polarity and number of peaks that they contain, the latency with which these peaks occur, and the distribution of ERP activity across the scalp (Kotz & Friederici, 2003). The exceptional temporal resolution provided by ERP measurement allows researchers to analyse very specific aspects of neurological functioning. This makes it ideal for studying language processing, which is founded on such a complex array of underlying mechanisms. At the time of writing this review, there are very few studies that have utilised ERP methods to investigate literacy development in children with any degree of hearing loss. In these, as well as in studies involving typically hearing children, the technique has been used to provide insight into the cognitive processes underlying normal and abnormal literacy development.

1.6.2. Semantic processing and the N400 effect. The 'N400' is an ERP component that is sensitive to the linguistic demands of a given EEG task. The waveform itself occurs predominantly in the central and parietal lobe regions, and is displayed as a negative peak at approximately 400 milliseconds after stimulus onset (Duncan et al., 2009; Kotz & Friederici, 2003). The amplitude of the N400 is especially sensitive to the predictability of linguistic context. More specifically, the extent to which word retrieval is facilitated by lexical or semantic factors may be identified as a clear reduction in waveform amplitude (Kutas & Federmeier, 2011). Hence, the difference in amplitude elicited by responses to semantically predictable and unpredictable stimuli is termed the 'N400 effect', and is used to index skills in semantic retrieval and lexical-semantic integration (Kotz & Friederici, 2003). The N400 effect has been successfully elicited in children as young as 5 years, although its topographical distribution at this age is comparatively broad (Byrne et al., 1999).

Evidence in support of the theorised link between the N400 effect and underlying semantic processing abilities comes from studies that elicit deviant electrophysiological responses in language-impaired populations. In one such study, 7- to 15-year-old children who were either typically developing or diagnosed with a specific language impairment (SLI) participated in a picture-word matching task (Cummings & Ceponiene, 2010). In this paradigm, participants are presented with a picture (e.g., a cloud), followed by either a matching (e.g., 'cloud') or non-matching (e.g., 'tree') auditory word stimulus. For both groups, the N400 elicited by non-matching stimuli was more negative-going than that elicited by matching stimuli, which indicated the existence of a N400 effect regardless of language ability. However, although the size of the N400 effect was similar between groups, the peak latency was significantly delayed in children with SLI. The authors suggested that this finding was representative of this group's verbal semantic integration deficits.

In another study comparing 'good' and 'poor' adult comprehenders, Landi and Perfetti (2007) used a similar task design to that employed by Cummings and Ceponiene (2010). Picture-picture and word-word pairs were presented consecutively on a computer screen, and were either unrelated (e.g., bran-teeth), categorically related (e.g., red-green), or both associatively and categorically related (e.g., mug-glass). Participants were asked to indicate whether the two items were similar in meaning or not. Again, both groups showed significant N400 effects. However, in the word pair condition, the mean amplitude of the N400 effect was significantly smaller in adults with poor oral comprehension abilities. As the groups were matched on nonword decoding and nonverbal IQ scores, these findings provide support for the theory that semantic processing is indexed by the N400 effect, and may present as surface-level skills in oral comprehension.

Some studies have also uncovered deviant N400 responses in populations with dyslexia. Schulz and colleagues (2009) found the mean amplitude of the N400 effect in 11-year-old children with dyslexia was significantly smaller than for typically developing children the same age. Electrophysiological responses were elicited by a sentence reading task, in which the final word in the sentence was semantically congruent or incongruent with the preceding sentence context (e.g., ‘The sky is [blue] / [fat]’). In contrast, Helenius, Salmelin, Service and Connolly (1999) found a similar mean amplitude of N400 effect in adults with and without dyslexia, although the peak of this effect occurred significantly later in dyslexic adults. Somewhat paradoxically, MEG results from the same study by Helenius et al. (1999) revealed that the amplitude of the N400 effect was in fact reduced in adults with dyslexia. Given that MEG techniques allow for higher spatial resolution and increased source localisation, the authors reasoned that there was indeed a difference between groups, and that this reflected suboptimal engagement of neural systems responsible for reading comprehension in dyslexic adults.

Another valuable source of evidence linking the N400 effect with underlying semantic processing is that provided by correlational analyses. Henderson, Baseler, Clarke, Watson and Snowling (2011) investigated the relationships between characteristics of the N400 component and scores on listening comprehension, vocabulary, and single word reading measures. In a sample of 8- to 10-year-old children, they found that N400 effect size, as indexed by the peak amplitude difference between congruent and incongruent picture-word pairs, was significantly and moderately correlated with behavioural measures of listening comprehension. Surprisingly, they did not find the same link with vocabulary scores. The authors suggested that this latter finding was explained by differences in the behavioural and EEG task demands, as performance on the behavioural measure gauges off-line processing, and requires other pragmatic, syntactic, and decision-making skills in addition to semantic retrieval and lexical-semantic integration (Henderson et al., 2011). Furthermore, they posited that the positive correlation between N400 effect amplitude and listening comprehension provided support for the N400 effect as an index of the ease with which semantic stimuli are integrated into context to achieve understanding.

1.6.2.1. N400 effect in populations with CIs. The N400 effect has been examined in numerous special populations, including those diagnosed with SLI (Cummings & Ceponiene, 2010), dyslexia (Helenius et al., 1999; Schulz et al., 2009), generalised learning disabilities (Fernandez, Silva-Pereyra, Prieto-Corona, Rodriguez-Camacho & Reynoso-Alcantara, 2014), autism spectrum disorder (McCleery et al., 2010) and William’s syndrome (Pinheiro, Galdo-Álvarez, Sampaio, Niznikiewicz & Gonçalves, 2010). Some studies have also been conducted to explore the N400 effect in individuals with CIs (Hahne, Wolf, Müller, Mürbe &

Friederici, 2012; Kallioinen et al., 2016; Key, Porter & Bradham, 2010; Vavatzanidis, Burbe, Friederici & Hahne, 2018). Such electrophysiological evidence has provided insight into the semantic processing capabilities of this population, thereby complementing behavioural evidence that highlights their spoken language difficulties (e.g., Geers, 2003; Nittrouer et al., 2012; 2014; Weisi et al., 2013).

Hahne and colleagues (2012) examined the N400 effect in adults with post-lingual hearing loss and CIs. A cloze sentence task was used, whereby the final word in an aurally presented sentence was either semantically congruent or incongruent with the preceding context. Between 300 and 700 milliseconds post-target onset, a significant N400 effect was elicited in adults with CIs and adults with typical hearing. For the group with CIs, this effect was also significant between 700 and 900 post-target onset, whereas this was not the case for adults with typical hearing. The longer-lasting nature of the N400 effect in adults with CIs was thought to represent delayed semantic integration processes (Hahne et al., 2012).

Compared with the research involving adults with CIs, there have been more studies investigating the N400 effect in children with CIs, although the results are arguably less conclusive. Kallioinen et al. (2016) used a word-picture matching paradigm to elicit ERP responses in 5- to 7-year-old children with CIs. The task included three different conditions: stimulus pairs were either congruent (e.g., wolf-wolf), between-category incongruent (e.g., wolf-car), or within-category incongruent (e.g., wolf-bear). A significant N400 effect in response to both types of incongruent stimuli was evident for CI children, as well as control groups of typically hearing children and children with hearing aids. Moreover, while the mean amplitude of the N400 effect elicited by within-category incongruence was similar across groups, the mean amplitude elicited by between-category incongruence was significantly greater for the CI group than for the other groups. This unexpected result was attributed to different processing strategies between groups. Since only a third of the total stimulus pairs were matching, the control group children may have realised that the target was not predicted by its prime in the majority of trials, thereafter taking a more passive approach to the task that reduced ERP effects associated with semantic relatedness. In contrast, children with CIs, who appeared to find the task more challenging, may have continued to use a predictive processing strategy that had, comparatively speaking, no effect on the size of the N400 effect. Further research is warranted to replicate and expand on these results, with consideration given to controlling potential strategy-related confounds.

N400 effects have also been reported to exist in younger, pre-school aged children with CIs. In a study by Vavatzinidis et al. (2018), participants aged between 2 and 6 years old passively viewed congruent and incongruent picture-word pairs. The group showed a significant N400 effect at 12, 18 and 24 months post-cochlear implantation. No baseline (i.e., pre-implantation) testing took place, which limits the degree to which results can be attributed to the use of hearing technology. There was also no typically hearing control group that would have enabled a direct comparison with age-based expectations. Nonetheless, the results indicate that young children with CIs may demonstrate a significant N400 effect. Similarly, in a case study reported by Key, Porter and Bradham (2010), a 6-year-old child with bilateral profound hearing loss demonstrated a visible N400 effect four months after receiving her second CI. Interestingly, the effect was absent prior to the activation of this CI and even two months post-activation, during which time she completed the picture-word passive viewing task with only unilateral auditory stimulation. Again, these results suggest – though not conclusively, given the single case sample size – that children with CIs can demonstrate sensitivity to semantic incongruence, as indexed by the N400 effect.

1.6.3. Phonological processing and the rhyme effect. Rhyme awareness starts to develop during the preschool years, as one of the earliest indicators of phonological sensitivity (de Jong & van der Leij, 2003; Ziegler & Goswami, 2005). A number of studies examining how phonological processing may be represented through electrophysiological measures have reported the existence of an ERP 'rhyme effect'; that is, the presentation of a non-rhyming word pair consistently elicits a larger, more negative-going ERP component than a rhyming word pair (Coch, Grossi, Skendzel & Neville, 2005). The particular component indexing this effect, referred to sometimes as the N450, is distributed across midline and right temporo-parietal regions of the scalp and peaks between 300 and 600 milliseconds post stimulus onset (Grossi, Coch, Coffey-Corina, Holcomb & Neville, 2001).

Given that reading difficulties are commonly attributed to a core deficit in phonological processing, it is perhaps unsurprising that some research involving the rhyme effect has revealed abnormalities in electrophysiological responses elicited in populations with dyslexia. Within such studies, ERP measurements are often recorded while participants perform a rhyme judgement task, wherein they rapidly decide the rhyming status of two stimuli presented consecutively on a screen. Noordenbos, Segers, Wagensveld and Verhoeven (2013) used this paradigm with first-grade children, who were grouped according to whether or not they were at genetic risk of dyslexia. Both groups, regardless of 'at-risk' status, showed significantly more negative-going rhyme effect mean amplitudes for non-rhyming stimuli, thus providing evidence of similar degrees of sensitivity to phonological manipulation. In contrast, Ackerman, Dykman and Oglesby (1994) reported that a sample of 7- to 12-year-old children with dyslexia showed no significant rhyme effect when presented with real word stimuli, and although they did show evidence of a rhyme effect when presented with nonword stimuli, it was significantly attenuated compared with groups of age-matched 'slow learners' and children with attention-deficit disorder.

The conflicting results reported by Noordenbos and colleagues (2013) may be due to differences in the severity of reading difficulties. For instance, although these children did perform significantly below controls on single-item reading measures, they were not yet diagnosed with dyslexia. Alternatively, the use of real word stimuli may have influenced children's neural responses to the task. As described in Section 1.6.2 of this literature review, the 'N400 effect' refers to the modulating effects of semantic relatedness on ERP waveform amplitude. The inclusion of real word stimuli in a task that measures phonological incongruence therefore makes the interpretation of findings challenging (Coch et al., 2005).

Lexical-semantic confounds aside, there is evidence to suggest that the rhyme effect relates to behavioural measures of phonological processing. In a study by McPherson, Ackerman, Holcomb and Dykman (1998), adolescents with dyslexia were divided into two groups on the basis of their scores on a behavioural nonword reading task. Based on mean rhyme effect amplitude differences elicited by a picture rhyme judgement task, dyslexic adolescents with poor nonword reading exhibited a significantly reduced rhyme effect, compared with both controls and other dyslexic adolescents. Importantly, these findings show that rhyme sensitivity, as indexed by the rhyme effect, may manifest specifically as nonword reading performance, for which accuracy relies fundamentally on the application of phonological processing skills (Coltheart et al., 2001). Similarly, Coch and colleagues (2005) divided a sample of 6- to 9-year-old typically developing children into two groups, based on performances on behavioural measures of phonological awareness. The onset of the rhyme effect elicited by an auditory version of a nonword rhyme judgement task was delayed by approximately 80 milliseconds in the below-average group, compared with the above-average group. Based on these findings, it may be expected that participants with phonological processing difficulties will show a deviant rhyme effect, in terms of either amplitude or latency of electrophysiological response.

Until recently, evidence for the rhyme effect elicited by purely visual stimuli extended only to participants aged 7 years and above. This is likely due to the potentially confounding variable of reading ability. For beginning readers, the task of decoding visually presented word and nonword stimuli is not yet automatic, and it may therefore be too cognitively demanding to include as a pre-requisite for measuring underlying phonological ability (Coch, Mitra, George & Berger, 2011). In order to overcome this confound, Coch and colleagues (2011) used a letter rhyme paradigm to gauge the nature of the rhyme effect in 6- to 9-year-old children. The stimulus pair that was presented to participants consisted of either rhyming (e.g., 'B'/'G') or non-rhyming letters (e.g., 'B'/'M'). Participants' responses therefore relied on knowledge of letter names, rather than reading ability. A significant rhyme effect was observed in the children, as well as in a separate group of young adults. Moreover, there were no differences between groups in terms of the amplitude or latency of the observed effect. Of note is the finding that the children's rhyme effect did not correlate significantly with behavioural measures of single word and nonword reading accuracy. According to the authors, the group's electrophysiological response does not appear to have indexed the same phonological processes that were applied to the standardised behavioural tests.

To date, there is no research investigating the rhyme effect in children with CIs, or even in children with hearing loss more broadly. That said, there are two studies that have examined the effect in adults with congenital hearing loss. MacSweeney, Goswami and Neville (2013) visually presented rhyming and non-rhyming word pairs to nine adults with severe-to-profound hearing loss and asked them to judge the stimuli's rhyming status. In response to the task, the group demonstrated a significant rhyme effect, the mean amplitude and latency of which was comparable to a control group of typically hearing adults. In contrast with these findings however, Colin, Zuinen, Bayard and Leybaert (2013) failed to elicit a significant rhyme effect in ten adults with moderate-to-profound hearing loss, who were administered a rhyme judgement task containing pictured stimulus pairs. Possibly, the equivocal findings reported by MacSweeney et al. (2013) and Colin et al. (2013) are attributable to the type of task stimuli used, although given that typically hearing adult control groups produced a significant rhyme effect in both studies, this factor cannot explain the discrepancy entirely. In both studies too, stimuli were matched between conditions on semantic concreteness, imageability and orthographic similarity, which means the results are not likely to have been influenced substantially by lexical-semantic confounds. Further research is therefore needed to tease apart the variables that may contribute to elicitation of the rhyme effect in adults with significant hearing loss.

In summary, there is evidence that sensitivity to rhyme may be measured by analysing the difference in electrophysiological response elicited by rhyming and non-rhyming stimulus pairs. Moreover, this rhyme effect has been found to exist in children as young as 6 years old. The assumption that the rhyme effect indexes underlying phonological processing is supported by research that shows abnormal responses in older children and adults with deficits in reading and phonological awareness. To date, there have been very few studies examining the rhyme effect in populations with any degree of hearing loss, and none to the author's knowledge that relate specifically to children. This therefore presents a significant gap in research evidence, especially given the abundant behavioural findings that highlight phonological processing difficulties in this population group (e.g., Ambrose et al., 2012).

1.7. Rationale and Aims for the Current Study

A review of the current literature reveals a significant lack of homogeneity in the sample characteristics of children with hearing loss. This is, in some ways, a necessary by-product of the large number of demographic and audiological factors that exist for the deaf and hard-of-hearing population. Still, too often, conclusions as to the effects attributable to underlying independent variables are made ambiguous by the existence of other, overlooked factors. The challenge for researchers is therefore to adequately account for the influence of a range of factors within their investigations. The present study was limited to children with CIs and bilateral hearing loss, who received their implants before the age of 5 years, and who were exposed to early AVT intervention. In addition, participants were in the first three years of formal literacy instruction (i.e., Grades 1-3) at the time of testing. This age-related criterion was considered particularly important, since a predominant aim of the research was to examine the relationships that exist between literacy sub-skills, and these are seen to change as a function of age in children with typical hearing (Catts et al., 2005). Moreover, the results of CI children in the present thesis were compared with those of typically hearing children matched on age, gender and nonverbal IQ. The homogeneity of CI participants and the inclusion of a matched control group allowed for valid conclusions to be drawn regarding the language and literacy abilities of hearing-impaired children with CIs.

The study described in Chapter 2 was based on a psycholinguistic approach to gathering and interpreting reading performance data. The broad aim of the study was to comprehensively examine the reading skills and sub-skills of children with CIs, and to gain insight into the processes underlying their early reading achievement. At a text level of written language processing, and with reference to the Simple View of Reading model, reading comprehension was here viewed as the product of both single word reading accuracy and listening comprehension. Using regression modelling, the relative contributions of these two underlying skill areas to reading comprehension were examined, in order to identify any differences in the pathways through which written text processing was achieved in children with CIs. At a single word level of written language processing, reading accuracy was examined with regard to the contributions made by phonological, orthographic and semantic processing skills (see Section 1.2 for discussion of theoretical models on which the rationale for selecting these skills is based).

The focus of Chapter 2, as stated above, was reading development. In Chapter 3, a similar psycholinguistic approach guided the examination of the same cohort's *spelling* development. Here, the aim was to examine the overall spelling performance exhibited by children with CIs, and to investigate the underlying literacy sub-skills contributing to their spelling achievement. Spelling accuracy results were analysed with respect to both irregular and nonsense words, thereby allowing for an examination of how underlying processing difficulties manifest differently depending on word type. Regression analyses for children with and without CIs were again generated on the basis of existing theoretical single word processing models, in order to compare the relative contributions of underlying phonological and orthographic skills to word spelling accuracy. Spelling errors were also analysed, to determine whether the phonological plausibility of inaccurate words differentiated groups. The study described in Chapter 3 was the first to specifically examine spelling development in children with CIs who used solely spoken communication, with comparisons based on a typically hearing control group matched on chronological age. The results therefore provided valuable insight into the written language skills of this population group.

The behavioural findings from Chapters 2 and 3 were complemented by electrophysiological data, as described in Chapters 4 and 5. Currently, few studies have used EEG methods to investigate language processing in school-age children with any degree of hearing loss. Yet, such techniques have been used with typically developing and special populations to gather valuable on-line information about the functioning of cognitive-linguistic neural processes. The same ERP methodologies were employed in the present thesis, in order to acquire neurophysiological evidence pertaining to semantic and phonological processing. Here, the aims of the studies were to examine the neural mechanisms that underlie literacy development in children with CIs, and to relate these findings with behavioural measures of spoken and written language processing.

Chapter 2.

Reading Development in Children with Cochlear Implants: A Psycholinguistic Investigation

Prior studies have consistently found that children with cochlear implants show reading deficits, although the exact sources of such deficits are difficult to disentangle, due to heterogeneous participant samples and a lack of psycholinguistic-based research. The study presented in this chapter aims to draw links between observable reading behaviours and underlying reading-related sub-skills, with reference to existing theoretical models of word- and text-level reading.

Chapter 2 is a corrected version of an article submitted for journal publication[1], which is currently under revision. Minor changes have been made to formatting and methodological description, in order to maintain thesis continuity. A section on statistical power analyses has also been added to this chapter's 'Results' section. The authors' contributions to the original submitted article are detailed on the following page.

[1] Bell, N., Angwin, A.J., Wilson, W.J., & Arnott, W.L. (2018*). Reading development in children with cochlear implants who communicate via spoken language: a psycholinguistic investigation.* Manuscript under revision.

Contributor	Statement of contribution
Nicola Bell (Candidate)	Conception and design: 70% Data collection and analysis: 100% Interpretation: 80% Manuscript writing: 100% Manuscript revisions: 20%
Dr Anthony Angwin	Conception and design: 5% Interpretation: 5% Manuscript revisions: 30%
A/Prof Wayne Wilson	Conception and design: 5% Interpretation: 5% Manuscript revisions: 20%
Dr Wendy Arnott	Conception and design: 20% Interpretation: 10% Manuscript revisions: 30%

2.1. Abstract

This study sought to comprehensively examine the reading skills and sub-skills of children with cochlear implants (CIs), and gain insight into the processes underlying their early reading development. Fourteen 6- to 9-year old children with CIs were assessed on a range of reading and spoken language measures. Their performances were compared to a control group of 31 typically hearing (TH) children of the same chronological and mental age. Group differences were examined using t-tests and regression modelling. Children with CIs performed significantly worse than TH children on reading accuracy, phonological processing and spoken language tasks. The predominant predictor of reading comprehension was word reading accuracy for the CI group, and listening comprehension for the TH group. Word reading profiles were similar across groups, with orthographic and phonological processing skills both contributing significant variance. Children with CIs demonstrated more early reading difficulties than their TH peers. As predicted by the Simple View of Reading model, successful reading comprehension for all children related to skills in listening comprehension and word recognition. The CI group's increased reliance on word reading accuracy when comprehending written text may stem from reduced word recognition automaticity. Despite showing reduced reading accuracy, children with CIs appeared to draw on orthographic and phonological skills to a similar degree as TH children when reading words in isolation.

2.2. Introduction

Literacy skills in typically developing children are not acquired on the sole basis of exposure to written language, but rather as the result of continuous and explicit instruction during the first few years of formal schooling (National Reading Panel, 2000). Word reading strategies that are taught to emergent readers pertain to which letter sequences represent which speech sounds, and they are therefore founded on – and develop alongside – underlying phonological processes (McBride-Change, 1999). Much evidence exists to support the theory that in children with typical hearing, there is a causal link between phonological processing deficits and poor reading outcomes (de Jong & van der Leij, 2000; Hulme, Snowling, Caravolas & Carroll, 2005; Wagner, Torgesen & Rashotte, 1994). The same link is thought to account for literacy difficulties in children with hearing loss, and indeed a large body of research has shown that children with aided severe-to-profound hearing loss perform poorly on phonological processing measures (Ambrose, Fey & Eisenberg, 2012; James, Rajput, Brinton & Goswami, 2008; Lee, Yim & Sim, 2012; Nittrouer, Caldwell, Lowenstein, Tarr & Holloman, 2012; Nittrouer, Caldwell-Tarr, Sansom, Twersky & Lowenstein, 2014; Spencer & Tomblin, 2009; Weisi et al., 2013). With respect to the cause of phonological processing deficits, children with hearing loss receive degraded auditory stimuli, even if benefiting from hearing aids or cochlear implantation, which may therefore result in the development of less fine-grained speech sound representations (Nelson & Crumpton, 2015).

Cochlear implantation has gained momentum in the past few decades as a realistic means by which those with a significant hearing loss can access spoken communication. After receiving cochlear implants (CIs), children's language-learning trajectories are often enhanced, particularly when implantation occurs early in life (Leigh, Dettman & Dowell, 2016; Tait, Nikolopoulos & Lutman, 2007). Even so, a discrepancy remains between the literacy outcomes of children with CIs and those of age-matched typically hearing (TH) children (Harris, 2015). The clarity of speech sound quality provided by CIs does not exactly imitate that provided by natural hearing (Ambrose et al., 2012; Nittrouer et al., 2012). Hence, early reading difficulties may still be expected to result from underlying limitations at the level of phonological processing. Adopting a psycholinguistic approach, the present study sought to further explore the impact of these phonological processing limitations on reading. To this end, the present study comprehensively examined word- and text-level reading skills and sub-skills, including phonological and orthographic processing and oral language, in beginning readers with CIs. The broad question addressed was whether, given observed deficits in key phonological processing skills, children with CIs achieved reading accuracy and comprehension on the basis of different underlying skills than similarly aged children with typical hearing. It was hypothesised that, in order to compensate for phonological difficulties, other underlying skills would contribute more to reading outcomes in children with CIs, when compared to TH children.

Abundant research has shown that children with CIs experience difficulties with reading comprehension (Geers, 2003; Nittrouer et al., 2012; 2014; Weisi et al., 2013), although given that the ability to draw meaning from a written text is dependent on a number of underlying skills, this finding alone cannot guide the provision of specific literacy support. According to the Language and Reading Consortium (2015), while the processes that contribute to reading comprehension are numerous and varied, their effects are enacted via the broad skill areas of word recognition (or word reading) and listening comprehension. These predictive relationships in reading comprehension are captured by the Simple View of Reading model (Catts, Adlof & Weismer, 2006; Hoover & Gough, 1990). An important aspect of the model is that the relative weightings of word reading and listening comprehension are age- and skill-dependent. The role of word reading in reading comprehension is expected to be most significant in early readers, for whom attentional resources and literacy instruction are devoted to decoding single words (Catts, Hogan & Adlof, 2005; Catts, Herrera, Nielson & Bridges, 2015; Language & Reading Consortium, 2015).

Past research has consistently demonstrated a significant relationship between word reading and reading comprehension in children with CIs (Geers, 2003; Johnson & Goswami, 2010; Vermeulen, van Bon, Schreuder, Knoors & Snik, 2007; von Muenster & Baker, 2014). Of these studies however, only two examined their cohort's performance in the context of concurrent spoken language performance and with reference to the Simple View of Reading model. Von Muenster and Baker (2014) reported that reading comprehension in children with CIs, aged 5 to 12 years, was strongly correlated with measures of word reading and receptive language. Similarly, Vermeulen and colleagues (2007) found that variance in reading comprehension scores for participants with CIs aged 7 years and older was significantly predicted by both word reading and receptive vocabulary scores. Based on these results, it may be speculated that children with CIs have a similar reading profile to that expected of their hearing peers, although importantly, the absence of a TH control group in each study undermines the certainty with which such a conclusion can be drawn. In addition, the broad age range of participants may mean that the changing predictive value of reading comprehension sub-skills over time is not effectively captured in the results.

A similar study by Nittrouer and colleagues (2012) examined the capacity of phonological awareness, executive functioning and oral language skills to predict reading comprehension in 6-year-old preschool children. For children with typical hearing, phonological and expressive vocabulary skills significantly predicted reading comprehension. In contrast, reading comprehension for children with CIs was only predicted by oral language measures (i.e., auditory comprehension, expressive vocabulary and narrative production). While these results point to a divergence in reading profiles between groups, potentially resulting from underlying phonological deficits in the children with CIs, there was no statistically significant group difference in the unadjusted weightings of predictor variables. Hence, the general profile of underlying skills on which CI children's reading comprehension is based may be similar to TH children. Further clarity on this point might be obtained if the research was extended to an older school-age group of readers, in whom reading skills would be better established. Given that phonological awareness has only an indirect effect on reading comprehension, examination of the primary mediatory factor in this relationship, single word reading, is also warranted.

Where the relationship between phonological awareness and word reading has been examined in early readers with severe-to-profound hearing loss, it is often found to be significant (Colin, Magnan, Ecalle & Leybaert, 2007; Harris & Beech, 1998; Nittrouer et al., 2012; von Muenster & Baker, 2014). However, there are also indications that other non-phonological skills may have a greater-than-expected influence in guiding the development of word reading ability. In the aforementioned study by Nittrouer and colleagues (2012), reading accuracy was significantly predicted by syllable awareness and narrative expression, whereas in TH children it was significantly predicted only by phonemic awareness. Hence, different processing skills were presumed to underlie word reading development in each group (Nittrouer et al., 2012). Notably, word reading performance in this study was indexed by the number of words read correctly in a written passage, rather than in isolation, which means that readers' performances were likely influenced by higher level language skills (e.g., contextual inference; Cain, Oakhill & Lemmon, 2004). Further research that explores the degree to which different underlying skills contribute to *decontextualised* word reading accuracy is warranted, and thus became the primary focus of the present study.

For typically developing children, the facilitative role of phonological processing in early word reading development has been well established (Badian, 1998; Castles & Coltheart, 2004; Hulme et al., 2002; Wagner et al., 1994). That this relationship exists is unsurprising, since in order to decipher unfamiliar words, a reader must be able to convert the given sequence of letters to a corresponding phonemic structure (Coltheart, Rastle, Perry, Langdon & Ziegler, 2001; Seidenberg & McClelland, 1989). Such a procedure may be contrasted with how familiar words are recognised by practiced readers, because these types of words can be directly accessed 'by sight' as whole or partial orthographic representations. In particular, 'irregular' words with uncommon letter-sound correspondences (e.g., 'scissors') cannot be wholly deciphered by converting letter sequences to phonemes, which means recognition of these items relies more on underlying orthographic processing (Hagiliassis, Pratt & Johnston, 2006). Phonological difficulties, such as those commonly exhibited by children with severe-to-profound hearing loss, may therefore differentially affect word reading accuracy, such that unfamiliar nonsense words are read more accurately than irregular real words. In the present study, word reading was measured using regular, irregular and nonsense words. This allowed for a psycholinguistic analysis of the functioning of underlying processors, thereby extending on research that has not examined phonological and orthographic processing as concurrent predictors of word reading in children with CIs (e.g., Nittrouer et al., 2012).

In another related study, James, Rajput, Brinton and Goswami (2009) explored the skills contributing to single real word processing in children with CIs aged approximately 8;4 years. On average, receptive vocabulary accounted for 45% of variance in word reading accuracy, whereas this value was only 18% for age-matched TH children. For another group of younger, reading-matched controls, rhyme awareness was the only significant predictor of word reading (45%), whereas this predictor for the CI group was much weaker (21%). Hence, the younger TH group appeared to be largely reliant on phonological awareness to achieve the same degree of reading accuracy as was attained by children with CIs, who were seemingly more reliant on vocabulary. Importantly, the children with CIs were exposed to auditory stimuli via the implants at a mean age of 4;7 years, which is quite late in life by current standards. In addition, a large proportion of the cohort used both spoken and sign language to communicate. It is unclear whether these factors influenced results, given that better literacy outcomes have previously been associated with early cochlear implantation (Ching, Dillon, Leigh & Cupples, 2017; Johnson & Goswami, 2010) and use of spoken language to communicate (Cupples et al., 2017; Johnson & Goswami, 2010; O'Donoghue, Nikolopoulos & Archbold, 2000). The present study therefore narrows the focus of investigation to a comparatively homogeneous CI cohort, whose members all received their implants earlier, used spoken communication, and were exposed to early auditory-verbal therapy (AVT) intervention.

Auditory-verbal therapy targets spoken communication development, and it is delivered to children with hearing loss in order that they might obtain age-appropriate speech and language outcomes (White & Brennan-Jones, 2014). The strong emphasis on spoken language in this approach is thought to promote emergent literacy skills by drawing explicit attention to the phonological constituents of speech (Kaderavek & Pakulski, 2007). Some studies have found that exposure to AVT is associated with better speech and language outcomes, relative to other interventions (Dettman, Wall, Constantinescu & Dowell, 2013; Percy-Smith et al., 2018). The only studies to have investigated *written* language proficiency in AVT-exposed children have done so without reference to an age-matched control group (e.g., Dornan, Hickson, Murdoch, Houston & Constantinescu, 2010; von Muenster & Baker, 2014). It is therefore not yet fully understood how the combined effects of AVT and cochlear implantation are reflected in the context of developmental, age-based expectations.

2.2.1. Current study. A review of recent literature reveals a substantial amount of heterogeneity in the characteristics of research participants with CIs. This variability is an almost inevitable by-product of the large number of demographic and audiological factors that exist for the hearing-impaired population at large. The challenge for researchers is therefore to adequately account for the influence of significant factors within their investigations. The present study's cohort of children with CIs were spoken language users who received AVT from an early age. Hence, it was not likely that communication mode would influence CI participants' performances differentially. Age at testing is another potential determinant of results: since relationships between literacy sub-skills may be expected to evolve over time (Catts et al., 2005), developmental changes may be obscured if analysed across a large age range. As such, the present study included only beginning readers, defined as children in the first three years of their formal literacy schooling. The first research question addressed in the present study was: How do the reading skills of children with CIs, who have received AVT, compare with those of TH children of the same age? Based on prior research involving similar clinical populations (e.g., Nittrouer et al., 2014; Weisi et al., 2013), it was hypothesised that children with CIs would perform significantly worse than TH children on measures of text-level and word-level reading, spoken language and phonological processing.

Previous study findings have indicated that the reading profiles of children with CIs may diverge from their TH peers, such that phonological processing deficits lead to an increased reliance on other skills like general spoken language (Mayberry, del Giudice & Lieberman, 2011; Nittrouer et al., 2012). Few studies, however, have analysed these relationships with systematic and explicit reference to current theoretical models of written language processing. Based on the Simple View of Reading model (Hoover & Gough, 1990), the second area of investigation in this study examined the relationship between reading comprehension and the two skill areas of which reading comprehension is composed: Do word reading and listening comprehension predict reading comprehension differently in children with CIs and TH children of the same age? It was predicted that reading comprehension profiles would not appear different between groups, and that better reading comprehension outcomes would be related to better word reading and listening comprehension performances (as was reported by von Muenster and Baker, 2014).

Finally, the word reading skills of the CI cohort in this study were examined from a psycholinguistic perspective, in order to determine the relative influence of underlying processing skills. Past studies involving typically developing children have identified phonological awareness as a critical facilitator of word-level literacy development, and indeed this has been the focus of many studies involving children with CIs. The current study extends prior research by placing the relationship between reading and phonology in the context of relationships between reading and other component skills (i.e., orthographic processing and vocabulary). We asked: Do phonological awareness, orthographic processing and vocabulary predict word reading accuracy differently in children with CIs and TH children of the same age? It was hypothesised that phonological awareness would predict concurrent word reading performance for both groups. Given the suggestion in previous research that processing of lexical information facilitates word recognition in CI children with reduced phonological processing skills (Geers & Hayes, 2010; Nittrouer et al., 2014), it was also hypothesised that the comparative contribution of non-phonological skills to word reading performance would be greater for the CI group.

2.3. Method

2.3.1. Participants. Fourteen children with CIs (7 females, 3 left-handed) and 31 TH children (16 females, 3 left-handed) participated in the present study. Most children with CIs and many of the typically hearing participants were recruited from Hear and Say, an organisation that provides audiological and spoken language intervention to children with hearing loss, as well as conducting regular school-based hearing screening. Parents and caregivers who had given permission to be contacted for research purposes were invited to participate in the study. The remaining participants were recruited from the wider community via newsletter advertisements and word of mouth.

All participants adhered to the following inclusion criteria: (1) use of spoken English as native and primary form of communication; (2) nonverbal reasoning at or above normal limits (as measured using the *Raven's Coloured Progressive Matrices*; Cotton et al., 2005; Raven, Raven & Court, 2004); (3) no diagnosed developmental disorders or intellectual disabilities, and; (4) in Grade 1, 2 or 3 of a mainstream school. The CI and TH groups did not differ on gender, $\chi^2(1) = 0.010$, $p = 0.920$, handedness, $\chi^2(1) = 1.153$, $p = 0.283$, age, $t(43) = -1.483$, $p = 0.145$, grade, $\chi^2(1) = 1.539$, $p = 0.463$, or nonverbal reasoning, $t(43) = 0.664$, $p = 0.510$. With regard to nonverbal reasoning, 'normal limits' was operationally defined as performance within one standard deviation of the mean (i.e., a z-score on the *Raven's Coloured Progressive Matrices* of between -1 and +1). Age and nonverbal reasoning scores were checked to confirm that data were normally distributed within each group and variances were equal between groups. There were no outliers for age or nonverbal reasoning in either group.

Additional inclusion criteria for the children with CIs were also implemented: participants needed to have received their CIs before the age of 5 years, and have received AVT before entering formal schooling. In all cases, AVT was administered by AVT-certified speech pathologists or AVT-certified Teachers of the Deaf. In AVT, speech and language goals are targeted through spoken communication. Families enrolled in the program generally attend both individual and group sessions. The frequency of individual sessions depends on the child's progress, so that when he or she has obtained test results within normal limits on two consecutive assessment points, they move from weekly to fortnightly appointments. Depending on progress, some children may move to monthly appointments, prior to their leaving the program at the commencement of formal schooling. The mean age at enrolment in early AVT intervention was 9 months (ranging from 0.08y to 3.25y).

The mean age of the CI group at the time of testing was 8.01 years (SD = 0.82y). These participants were in Grade 1 (n = 1), Grade 2 (n = 6) or Grade 3 (n = 7) at the time of testing. For context, in the Australian state of Queensland (in which all participants attended school), children entering Grade 1 are 6 years old by July 1 of that year. The CI group's average nonverbal reasoning z-score was 0.72 (SD = 1.21) as measured using the *Raven's Coloured Progressive Matrices*. All participants in the group had bilateral sensorineural hearing loss, which was either stable (n = 10) or progressive (n = 4). Aided pure tone average (PTA) hearing thresholds for both ears were at or under 30dB for all children (see Table 2.1). Twelve participants from the group had bilateral CIs, while two had a unilateral implant and an additional hearing aid. Twelve participants used a Cochlear Nucleus 6 (CP910) model of processor, one used a Cochlear Nucleus 5 (CP810), and one used a Naida CIQ70. All children received CI surgery before the age of 5 years (mean = 1.74y; SD = 1.45y), and the mean duration of CI use at the first point of current testing was 6.23 years (SD = 1.52y). Participants were reported to use their CIs during all waking hours. Additional information pertaining to each participant is shown in Table 2.1.

Participants in the TH control group passed hearing screening assessments (thresholds of 25dB HL or better at octave intervals of 500 to 4000 Hz) conducted using a commercially available screening audiometer. The participants completed these screenings after entering formal schooling and no more than two years prior to their participation in the present study. The mean age of the TH group was 7.63 years (SD = 0.77y). Five participants were in Grade 1, 16 were in Grade 2, and 10 were in Grade 3 at the time of testing. The group's average nonverbal reasoning z-score on the *Raven's Coloured Progressive Matrices* was 0.95 (SD = 0.98).

2.3.2. Ethics statement. Ethical approval for the present study was obtained from the Behavioural and Social Sciences Ethical Review Committee at the University of Queensland. Gatekeeper ethical approval was also obtained from the Hear and Say Research and Ethical Advisory Committee. All parents gave written informed consent for their children to participate in the study.

Table 2.1

Audiometric information for participants with CIs (n=14).

#	Configuration	Aetiology	Stable (Y/N)	Age (m) at 1st implant	Age (m) at EI enrolment	Unaided PTA (dB)		Aided PTA (dB)	
						Left	Right	Left	Right
1	CI+CI	Con. (idiopathic)	Y	46	2	75.00	76.25	25.00	25.00
2	CI+CI	Con. (idiopathic)	Y	7	1	98.75	≥100	26.25	25.00
3	CI+CI	Con. (idiopathic)	Y	7	2	92.50	92.50	25.00	26.25
4	CI+CI	Con. (idiopathic)	N	37	29	75.00	63.75	22.50	22.50
5	CI+CI	Con. (idiopathic)	N	31	3	88.75	82.50	22.50	18.75
6	CI+CI	Con. (idiopathic; LVAS)	Y	9	4	76.25	97.50	21.25	22.50
7	CI+CI	Con. (genetic-NS)	Y	8	2	98.75	98.75	25.00	22.50
8	CI+CI	Con. (genetic-Connexin 26)	Y	7	1	≥100	≥100	22.50	18.75
9	CI+CI	Con. (genetic-Connexin 26)	Y	8	1	95.00	≥100	22.50	21.25
10	CI+CI	Acq. (1;3y; idiopathic)	Y	19	31	≥100	≥100	27.50	22.50
11	CI+CI	Con. (CMV)	Y	8	4	≥100	98.75	25.00	21.25
12	CI+CI	Con. (CMV)	Y	8	7	97.50	≥100	30.00	28.75
13	CI+HA	Con. (genetic-Pendred Syndrome)	N	47	1	96.25	75.00	28.75	*
14	HA+CI	Acq. (1;3y; idiopathic; LVAS)	N	50	39	61.25	77.50	30.00	30.00

Note. CI = cochlear implant; HA = hearing aid; EI = early intervention; PTA = pure tone average; Con. = congenital; Acq. = acquired; LVAS = Large Vestibular Aqueduct Syndrome; NS = not specified; CMV = Cytomegalovirus. *Data for left ear missing but reportedly similar to right ear.

2.3.3. Tests and materials.

2.3.3.1. Word-level reading. Word-level reading accuracy was assessed using the *Castles and Coltheart 2* (*CC2;* Castles et al., 2009*),* a standardised test normed for Australian children in Grades 1 through 6 (i.e., age 6 through 11;6 years). Examinees are asked to read aloud a combination of three different word types: (1) nonwords, which have regular spellings but are not real words (e.g., 'gop'); (2) irregular words, which have irregular English phoneme-grapheme correspondences (e.g., 'good'); (3) regular words, which have regular English phoneme-grapheme correspondences (e.g., 'bed'). In total, there are 120 items of increasing difficulty (40 of each word type), and each item is presented on an individual card. The examinee is scored 1 for an item if their response accurately reflects the standard adult pronunciation. A score out of 40 for each word type was obtained and this was converted to a standardised z-score, using the test norms.

2.3.3.2. Text-level reading. In addition to word-level reading tasks, participants' passage (or text-level) reading ability was assessed using the *Primary Passage* subtest of the *York Assessment of Reading for Comprehension – Australian edition* (*YARC*; Snowling et al., 2012). Performances were scored using the test's standardised Australian norms for children aged 5 to 12 years. In this assessment measure, children read aloud two passages of text selected according to their age and accuracy level. They then answer approximately eight questions about the passage, which require use of both literal and inferential comprehension skills. Scores are calculated for reading comprehension (i.e., cumulated '0' or '1' scores, depending on accuracy with answering questions), reading rate (i.e., the time taken to read aloud the given text), and reading accuracy (i.e., the total number of errors observed while reading). Raw scores for reading comprehension, rate and accuracy were converted to standard scores using the procedure outlined in the test manual.

2.3.3.3. Reading sub-skills.

2.3.3.3.1. Phonological awareness. Three subtests from the *Comprehensive Test of Phonological Processing – 2nd edition* (*CTOPP-2*; Wagner, Torgesen, Rashotte & Pearson, 2013) were administered to assess phonological awareness. The CTOPP-2 is a norm-referenced test, with standardised scores available for children aged 4 and over. An index score representing phonological awareness skill was calculated on the basis of children's performance on: (1) *Elision*; (2) *Blending Words*; (3) *Sound Matching* (4-7 years), and; (4) *Phoneme Isolation* (7+ years). In subtest (1), examinees were asked to produce a given word with one phoneme omitted (e.g., 'What is "flame" without /f/?"). In subtest (2), examinees were given isolated spoken word parts and asked to blend them together to form a word (e.g., 'What is /s/ - /t/ - /æ/ - /m/ - /p/?'. In subtest (3), examinees (4-7y) were asked to match a pictured target word with one of three or four other pictured words, based on a common phoneme (e.g., 'Which of these words has the same first sound as "cat": "rug", "car", or "mat"?'). In subtest (4), examinees (7y+) were asked to identify the target phoneme from a spoken word (e.g., 'What is the third sound in "frog"?'). As outlined in the test manual, raw scores from each of these subtests were combined and converted to a composite phonological awareness score.

2.3.3.3.2. Rapid automatised naming. The CTOPP-2 subtests designed to assess symbolic rapid automatised naming (RAN) were also administered. Rapid automatised naming is described by the test authors as a measure of phonological retrieval. Examinees are asked to quickly name symbolic items (e.g., digits or letters) in an array. It is not a test of accuracy, but of the speed or efficiency with which the stored label for each item is accessed and pronounced.

2.3.3.3.3. Phonological memory. Phonological memory was assessed using the *Children's Test of Nonword Repetition* (*CNRep*; Gathercole & Baddeley, 1996). Standardised scores and percentile ranks are available for children aged 4;0 to 9;11 years. In this test, children are asked to repeat a given nonword. There are 40 items on the test, divided equally into two-, three-, four- and five-syllable nonwords. Nonword repetition has been found to reflect phonological processing skills, and is a significant predictor of reading ability in children (Wagner et al., 1994).

2.3.3.3.4. Letter-sound knowledge. Letter-sound awareness is an important precursor to reading development (Catts et al., 2015). The *Letter-Sound Test* (*LeST*; Larsen, Kohnen, McArthur & Nickels, 2011) was used to measure children's explicit knowledge of phoneme-grapheme correspondences. Australian norms for children aged 5;0 to 9;11 years were used to convert raw scores to standardised z-scores (Larsen, Kohnen, Nickels & McArthur, 2015). In this task, examinees are visually presented with 51 individual letters (or letter groups like 'ch') and are asked to say aloud the associated sound. Items are scored either '0' (incorrect) or '1' (correct), and the total raw score can be converted to a standardised score.

2.3.3.3.5. Orthographic processing. Orthographic processing was assessed using the *Test of Orthographic Choice* (*TOC*; Kohnen, Anandakumar, McArthur & Castles, 2012).

This assessment, which has standardised norms for Grades 1 through 6, measures written word recognition. Examinees are presented with two words, both of which may be pronounced identically, but only one of which is a real word (e.g., 'caip'/'cape'). Children are asked to circle which of the items is a real word. The test contains 30 items and two practice items. Accuracy depends on retrieval of the word's orthographic representation, rather than knowledge of the word's meaning or the ability to 'sound it out'. Raw scores out of 30 were converted into standardised z-scores.

2.3.3.4. Oral language.

2.3.3.4.1. Receptive vocabulary. Receptive vocabulary was assessed using the *Peabody Picture Vocabulary Test – 4th edition* (*PPVT-IV*; Dunn & Dunn, 2007). Standardised norms are available for individuals over the age of 2;6 years. In the *PPVT-IV*, examinees are presented with four pictures and are asked to point to the one that corresponds with a word spoken by the test administrator. There are 228 items in total, composed of 19 equally distributed and increasingly difficult item-sets. The test is widely used as a measure of receptive vocabulary, or the ability to understand the meaning of single words in isolation. Raw scores were converted to standard scores.

2.3.3.4.2. Core language. The *Clinical Evaluation of Language Fundamentals – 4th edition* (*CELF-4*; Semel, Wiig & Secord, 2003) contains a comprehensive battery of oral language assessments. A core language index score, which is designed to represent overall expressive and receptive language ability, was calculated on the basis of: (1) *Concepts and Following Directions*; (2) *Word Structure*; (3) *Recalling Sentences*, and; (4) *Formulated Sentences.* In subtest (1), examinees are required to follow increasingly difficult spoken directions. In subtest (2), examinees use word endings as grammatical markers. In subtest (3), examinees repeat increasingly complex and lengthy sentences. In subtest (4), examinees construct their own sentences to describe a given picture. The core language index composite index score is derived from raw scores on each of these tests, and in the present study, was converted to a standard score for analyses.

2.3.3.4.3. Understanding spoken paragraphs. An additional supplementary subtest called 'Understanding Spoken Paragraphs' was also administered. The subtest requires the examinee to answer a number of questions pertaining to a piece of text that was read to them just prior. The format of this test is therefore analogous to that of the *YARC*, and so the administration of both allowed for comprehension difficulties to be categorised as having either spoken language or reading origins. Australian norms for this and other subtests of the *CELF-4* are available for children older than 5 years. The scaled score for this subtest was used in analyses.

2.3.4. Procedure. All participants were individually administered the aforementioned language and literacy assessments in a quiet room. Testing took place over the course of two or three sessions (approximately three hours in total). The number of sessions varied between participants and depended upon time constraints and children's level of fatigue. The standardised protocols were adhered to, and all assessments were administered by the same qualified speech pathologist. Where possible, all participants received the tests in the same order, although some variation occurred depending on participant temperament and availability of testing materials. During the sessions, test breaks were permitted whenever requested by the child, or whenever deemed necessary by the examiner. Assessments were scored during the testing session where possible. Participants' responses to the tasks were also aurally recorded using a Philips Voicetracer 620, so that any items that were missed or unclear could be verified at a later time by the same test administrator.

2.4. Results

2.4.1. Group comparisons. The first research question pertained to the reading performances of children with CIs, compared with the TH cohort. In all analyses, standardised scores for each test were the dependent variables of interest. One-tailed independent samples t-tests were conducted with measures on which it was hypothesised children with CIs would perform worse than children with typical hearing (i.e., all reading, spoken language and phonological processing tasks). In contrast, a two-tailed t-test was used for the measure of orthographic processing since the scarcity of literature in this area made predicting the direction of effect uncertain. Histograms for each dependent variable were examined separately for each group to confirm that the assumption of normality was satisfied, and Levene's test for equality of variances was used to further confirm that this assumption was met.

Results of statistical comparisons for the CI and TH groups are summarised in Table 2.2. In terms of reading performance (Figure 2.1), the CI group obtained significantly worse scores than the TH group on text reading accuracy (p = 0.009), with a medium effect size (i.e., Cohen's d). Children with CIs also scored worse than the TH group on reading comprehension, although this did not reach statistical significance (p = 0.072). The difference between groups on reading rate was non-significant. For single word reading accuracy, the CI group's score was significantly lower than the TH group for regular words ($p = 0.036$) and nonwords ($p = 0.025$), again with medium effect sizes for each word type. The group difference for irregular word reading accuracy was not significant ($p = 0.054$).

Analyses of underlying literacy sub-skills showed that the CI group performed worse on measures of phonological awareness ($p < 0.001$), phonological memory ($p < 0.001$), RAN ($p = 0.010$) and letter-sound knowledge ($p = 0.030$). Effect sizes ranged from medium (RAN, letter-sound knowledge) to large (phonological awareness, phonological memory). The CI group performed significantly worse than the TH group on spoken language measures of receptive vocabulary ($p = 0.005$) and overall spoken language ($p < 0.001$), and these effect sizes were large. There were no statistically significant differences between groups on orthographic processing and listening comprehension (as measured by the 'Understanding Spoken Paragraphs' test).

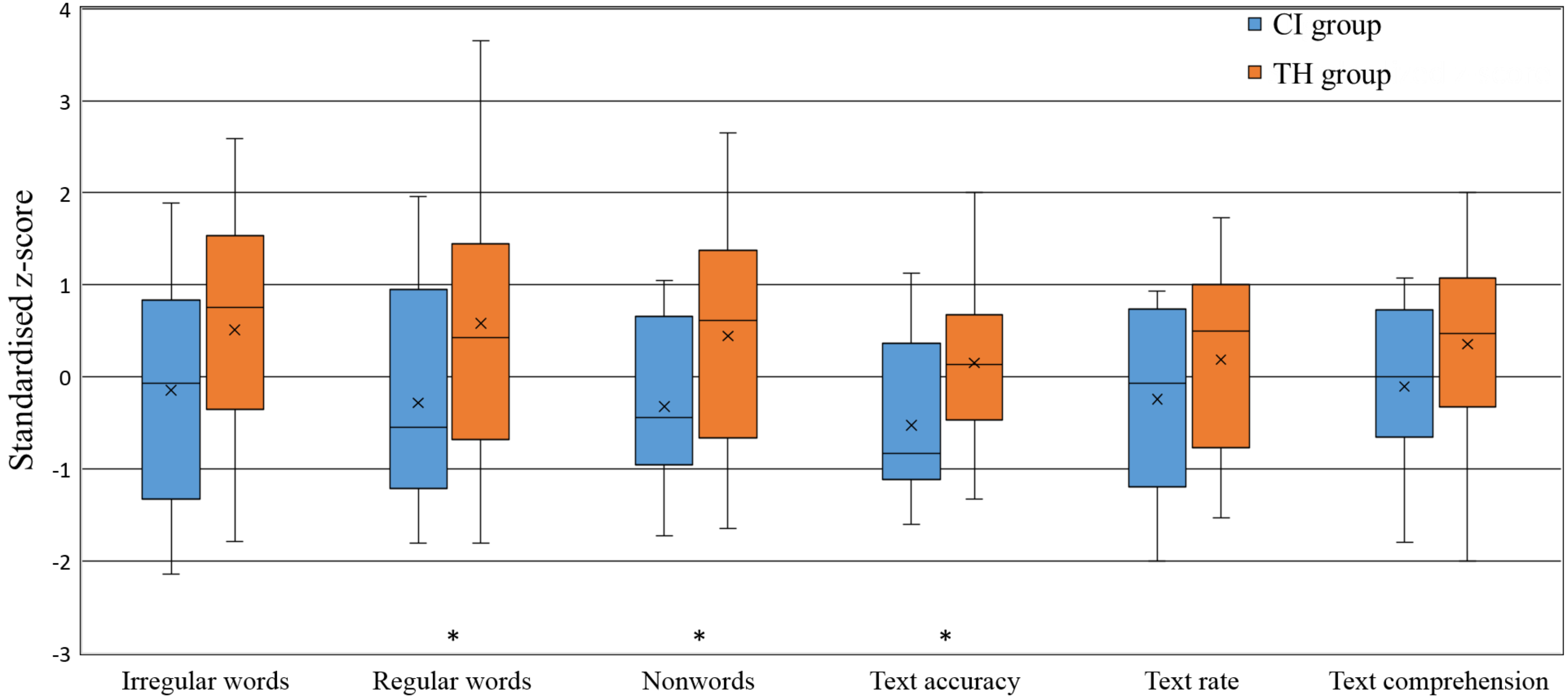

Figure 2.1. Box and whisker plots showing distribution of reading task performances by CI and TH groups.
Note. CI = cochlear implant; TH = typically hearing. Significantly poorer ($p<.05$) mean scores for CI group on regular word, nonword and text reading accuracy (as indicated by *). Whiskers above and below the box mark maximum and minimum values respectively; upper box boundary marks the 75^{th} percentile; lower box boundary marks the 25^{th} percentile; horizontal line within box boundary marks the median; cross (X) marks the mean.

Table 2.2

CI vs. TH group comparisons on all language and reading measures.

Domain	**Measure**	**CI Mean (SD)**	**TH Mean (SD)[1]**	***t***	**Cohen's *d***	**[2]*p***
Word-level reading	Regular words	-0.28 (1.13)	0.58 (1.57)	1.84	.63	*.036
	Irregular words	-0.14 (1.29)	0.51 (1.20)	1.65	.52	.054
	Nonwords	-0.32 (0.91)	0.45 (1.27)	2.02	.70	*.025
	Word reading composite	-0.25 (1.08)	0.52 (1.26)	1.95	.66	*.029
Text-level reading	Reading accuracy	92.07 (13.24)	102.26 (12.85)	2.44	.78	*.009
	Reading rate	96.38 (16.03)	102.79 (15.05)	1.24	.41	.111
	Reading comprehension	98.43 (13.28)	105.32 (14.08)	1.49	.50	.072
Reading sub-skills	Phonological awareness	90.71 (13.48)	107.45 (14.51)	3.66	1.19	*<.001
	RAN	92.93 (10.28)	100.40 (9.08)	2.44	.77	*.010
	Phonological memory	-1.33 (1.02)	0.28 (0.88)	5.44	1.70	*<.001
	Letter-sound knowledge	-0.34 (1.13)	0.30 (0.97)	1.93	.61	*.030
	Orthographic processing	0.11 (1.25)	0.36 (1.22)	.62	.20	.541
Oral language	Receptive vocabulary	102.64 (17.75)	115.94 (14.24)	2.68	.81	*.005
	Core language	94.86 (12.98)	109.23 (11.95)	3.49	1.15	*<.001
	Understanding Spoken Paragraphs	9.13 (2.56)	9.74 (2.35)	.95	.25	.174

Note. CI = cochlear implant; TH = typically hearing; RAN = rapid automatised naming. Statistically significant values ($p<.05$) indicated with asterisk (*). [1]Missing values for one TH participant on word-level reading, RAN and letter-sound knowledge (i.e., n=30 for these reported values). [2]1-tailed *p*-values reported for all tests except orthographic processing.

2.4.2. Text-level reading relationships. The second research question in the present study asked whether reading comprehension was based on a different profile of underlying skills between the two groups of CI and TH children. The outcome measure of reading comprehension was represented by participants' reading comprehension standard scores on the *YARC*. Word reading was represented by a word reading composite score, calculated as the average of each participant's regular word, irregular word and nonword reading accuracy standard scores on the *CC2*. Listening comprehension was represented by participants' scaled scores on the *CELF-4* 'Understanding Spoken Paragraphs' subtest. Word recognition and listening comprehension were first entered individually (and separately for each group) as predictor variables in univariate linear regression analyses. In each case, reading comprehension was the outcome variable. To examine the value of each predictor variable when entered simultaneously in the one regression model, multiple regression analyses were then conducted for each group.

The unadjusted and adjusted regression analysis outcomes (summarised in Table 2.3) revealed that reading comprehension was concurrently predicted by both listening comprehension and word reading accuracy for the two groups. For CI children, word reading was the predominant predictor, and for TH children listening comprehension was the predominant predictor.

Table 2.3

Multiple regression results for reading comprehension.

	CI				TH			
	R^2_{adj}	β	*t*	*p*	R^2_{adj}	β	*t*	*p*
Unadjusted								
WR	*.364	.650	2.961	*.012	*.379	.632	4.321	*<.001
LC	.201	.512	2.065	.061	*.534	.534	.742	*<.001
Adjusted	*.536				*.607			
WR		.593	3.109	*.010		.328	2.379	*.025
LC		.434	2.276	*.044		.571	4.150	*<.001

Note. CI = cochlear implant; TH = typically hearing; WR = word reading composite on *Castles and Coltheart 2*; LC = listening comprehension on *Understanding Spoken Paragraphs* subtest of *Clinical Evaluation of Language Fundamental 4th ed*. Statistically significant values (p<.05) indicated with asterisk (*).

2.4.3. Word-level reading relationships. Overall word reading accuracy was significantly poorer for children with CIs in the current study, as shown in Table 2.2. The third research question pertained to the sub-skills presumed to underlie this outcome. Specifically, the dependent variable of interest was word reading accuracy (again represented by a *CC2* word reading composite), with predictor variables of interest being phonological awareness, orthographic processing and semantic processing. Phonological awareness (PA) was represented by the composite standard score on the *CTOPP-2*. Orthographic processing (OP) was represented by the standard (z-) score on the *TOC*. Semantic processing was represented by participants' receptive vocabulary (RV; *PPVT-4*) standard scores.

Relationships between these sub-skills and overall word reading accuracy were first analysed with univariate regression analyses. As shown in Table 2.4, the unadjusted regression coefficients for each predictor variable were statistically significant, within both the CI and TH groups. Multiple regression analyses were then conducted, in order to examine predictors' values when adjusted for the simultaneous contributions of other predictors (i.e., when PA, OP and RV were included in the one regression model).

For both groups, RV did not contribute any significant unique variance to word reading performance beyond what was contributed by PA and OP (data not shown). In light of this finding, and due also to concerns about collinearity between predictor variables, RV was subsequently removed as a predictor from the model. The resultant regression model, containing only PA and OP, is shown in Table 2.4. The word reading profiles of CI and TH groups were similar, in that OP contributed the most variance overall to word reading performance, while PA contributed less so, but still significantly.

2.4.4. Power analyses. A post hoc analysis was carried out with the CI group, in order to determine the statistical power available for multiple regression analyses with 14 participants and two predictor variables. Results indicated that the power value required to find a squared multiple correlation coefficient of medium size (i.e., 0.5) was 0.65. This value was considered acceptable, although a limitation of the study was that there may have been insufficient power to find smaller multiple correlation coefficient values. Despite the small sample size however, the text- and word-level regression analyses described in Sections 2.4.2 and 2.4.3 did return significant results for the CI group, indicating that the relationships examined were strong, and that a Type II error was not made in this case.

Table 2.4

Multiple regression results for word reading accuracy.

	CI				TH			
	R^2_{adj}	β	*t*	*p*	R^2_{adj}	β	*t*	*p*
Unadjusted								
OP	*.670	.834	5.233	*<.001	*.529	.738	5.790	*<.001
RV	*.318	.608	2.655	*.021	*.367	.623	4.217	*<.001
PA	*.347	.631	2.814	*.016	*.305	.573	3.702	*.001
Adjusted	*.819				*.618			
OP		.706	5.675	*<.001		.608	4.899	*<.001
PA		.410	3.294	*.006		.341	2.751	*.010

Note. CI = cochlear implant; TH = typically hearing; OP = orthographic processing on *Test of Orthographic Choice*; RV = receptive vocabulary on *Peabody Picture Vocabulary Test 4th ed.*; PA = phonological awareness composite score on *Comprehensive Test of Phonological Processing 2*. Statistically significant values (p<.05) indicated with asterisk (*).

2.5. Discussion

The aims of the current study were to investigate the reading skills of beginning readers with CIs, with specific regard to: (1) how they performed in comparison to TH children of the same age; (2) the skills underlying their reading comprehension, and; (3) the skills underlying their word reading accuracy. As predicted, there were key differences between the CI and TH groups in terms of their written and spoken language abilities. Reading comprehension appeared to be based on similar underlying skills for all children, though to different degrees between groups. An increased reliance on word reading accuracy observed for children with CIs may reflect a delay in word processing automaticity. Word-level reading accuracy related to orthographic and phonological processing skills for both groups, although despite this resemblance in profiles, children with CIs obtained worse outcomes. Implications of the above findings are discussed in detail below.

2.5.1. Reading and language in beginning readers with cochlear implants. In comparison to their TH peers, children with CIs in the present study had significantly worse overall single word reading accuracy. Previous authors have reported similar findings with cohorts of school-age children with CIs (e.g., Harris & Terlektsi, 2011; Nittrouer et al., 2014; Spencer & Tomblin, 2009; Weisi et al., 2013). This study extends those results by focusing on children who are homogeneous in age at assessment, consistent use of CIs and spoken communication, prior exposure to AVT, an absence of additional disabilities, and normal nonverbal reasoning. Additionally, word reading stimuli were systematically manipulated in terms of lexicality (i.e., real and nonsense words) and regularity (i.e., regular and irregular real words). Children with CIs performed significantly worse than TH children when reading aloud nonwords and regular words, whereas the group difference associated with irregular word reading was only borderline significant.

The difference in reading accuracy between word types for the CI group may be explained by underlying phonological deficits. According to existing theoretical models, skilled readers draw on phonological, orthographic and semantic processes to recognise single words, though not to the same extent across word types (Coltheart et al., 2001; Seidenberg & McClelland, 1989). Nonwords are deciphered using knowledge of which letter sequences usually represent which phonemes. Irregular words, by definition, contain uncommon letter-sound correspondences, the orthographic representations of which are accessed at least partially from long-term storage (Hagiliassis et al., 2006). In the present study, children with CIs appeared to have more success (relative to their TH peers) reading aloud irregular words than nonwords, and thus the observed pattern of results suggests the children with CIs may have had specific difficulties applying phonemic 'decoding' skills when reading.

In support of the hypothesis for phonological processing deficits, the CI group performed significantly worse on tasks with high phonological processing demands. Phonological memory, as measured by a nonword repetition task, was particularly discrepant between groups. To complete this task, the examinee must recover phonological representations from an unfamiliar phonemic sequence presented aurally, store those representations in working memory, and then access this information to produce the sequence exactly (Gathercole, 2006). A breakdown in any one of these steps may therefore lead to imitation errors. In the present study, children with CIs obtained a mean nonword repetition score that was more than one standard deviation below the test's normative sample. Past studies have highlighted nonword repetition as an important predictor of reading proficiency in hearing children with dyslexia (Berninger et al., 2009) and language impairment (Dollaghan & Campbell, 1998; Weismer et al., 2000), and also in children with severe-to-profound hearing loss who use CIs (Casserly & Pisoni, 2013; Dillon & Pisoni, 2006; Edwards & Anderson, 2014; Nittrouer et al., 2014). Thus, the current study's findings align with the general pattern of results, highlighting phonological memory as a significant area of difficulty for this population.

Other skills pertaining to phonological processing, including phonological awareness, RAN and letter-sound knowledge, were also significantly poorer in the CI group. This pattern of results was unsurprising, given existing research in the area (Ambrose et al., 2012; James et al., 2008; Lee et al., 2012; Nittrouer et al., 2012; 2014; Spencer & Tomblin, 2009; Weisi et al., 2013). Such phonological deficits are presumed to reflect less fine-grained speech sound representations, which are founded on the comparatively degraded speech signal provided by CIs (Nittrouer et al., 2012). In contrast, there were no significant group differences in orthographic processing skills, which may therefore be a relative strength for children with CIs (Bouton, Colé, Serniclaes, Duncan, & Giraud, 2015).

Interestingly, while TH children still outperformed the CI cohort in reading comprehension, the difference between groups only approached statistical significance. This finding represents a better outcome for children with CIs relative to those reported in past studies (Geers, 2003; Nittrouer et al., 2014; Spencer, Barker & Tomblin, 2003; Weisi et al., 2013). The positive results in the present study may be attributable to the CI group's high nonverbal IQ (mean z-score = 0.72) and the early age at which they received their implants (mean age = 1.74y). Our analyses focused only on beginning readers in the first few years of formal schooling, so it may also be the case that reading comprehension difficulties emerge in later years, when the academic demands are such that these children are expected to 'read to learn', rather than 'learn to read'. Indeed, the trend in other studies is for the performance gap between children with CIs and children with typical hearing to widen with age (Geers & Hayes, 2010; Geers, Tobey, Moog & Brenner, 2008).

In terms of oral language, children with CIs in the present study had significantly poorer overall spoken language skills than their peers. These group differences were predicted, based on previous reports of difficulty in receptive vocabulary and receptive and expressive oral language in the CI population (Ceh, Bervinchak & Francis, 2013; Fitzpatrick et al., 2012). Here too, the present study's findings expand on past research involving children with hearing loss and early exposure to AVT. Fulcher, Purcell, Baker and Munro (2012) reported positive spoken language outcomes for 5-year-old children with varying severities of hearing loss who had received AVT, as reflected in the high proportion of 'within normal limits' performances. Unlike the present study, however, Fulcher et al. (2012) and other studies that have focused on AVT-exposed children (e.g., Dornan et al., 2010; von

Muenster & Baker, 2014) did not include a TH control group, with whom the AVT group's skills could be directly compared. Findings from the present study revealed no significant group difference between the CI and TH children in their ability to understand spoken paragraphs. The format of the test used to measure this skill was very similar to the reading comprehension measure, except that passage stimuli were presented aurally, rather than in print. The extent to which each group relied on listening comprehension skills to achieve text-level reading comprehension was examined using regression analyses, as is described in Section 2.5.2.

Typically, children's language skills develop on the basis of exposure to spoken vocabulary and syntactic structures (Kuhl, 2004). Spoken language difficulties experienced by individuals with CIs may therefore be attributed to degraded auditory exposure to such structures after implantation, since the auditory signal received via a CI is not as sensitive as in typical hearing. In addition, children with CIs in the present study had no or very little access to auditory stimulation via their affected ear(s), during the period of time when they had substantial hearing loss but did not yet have their CI(s). For those with congenital hearing loss, their first exposure to spoken language would have been at the point of CI 'switch-on', while for others with acquired or progressive hearing loss, there would have been a period before implantation during which they had access to some sound, either with or without a hearing aid. This variability in 'hearing age' (i.e., duration of aided exposure to spoken language) may be considered a limitation of the present study. Future research should address how differences in hearing age – or pre-CI hearing experiences more broadly – influence spoken and written language development in beginning readers with CIs.

It should be noted that for the current CI group average, receptive vocabulary performance exceeded the normative sample mean. In fact, only two of the 14 participants with CIs scored below normal limits. The significant difference was therefore largely driven by the high score achieved by the TH group. In all other measures except phonological memory, the CI group average was within normal limits (i.e., above 85 standard scores). Again, such a positive outcome for the CI group, relative to results reported elsewhere in the literature, may be attributable to the cohort's high average nonverbal reasoning score. Nonetheless, given that the CI and TH groups had similar nonverbal skills, persisting discrepancies in vocabulary and other spoken and written language performances suggest these skills may not be as high for children with CIs as would otherwise be expected, based on nonverbal cognitive abilities alone.

As well as being homogeneous in the type of early intervention received, participants in our CI group were also all spoken communicators and had ‘normal’ nonverbal reasoning skills. These individual factors have been found in past studies to influence language and literacy outcomes (Cupples et al., 2017; Geers, 2003; Harris & Beech, 1998), so were intentionally controlled to reduce the effects of potentially confounding variables. As such, the present results are not necessarily generalisable to children with hearing loss who do not meet the same criteria. Future studies may target similar questions as those addressed here, but with a larger, more heterogeneous sample, and thus a broader range of variables for input into statistical analyses. That said, there was some variability in the age at which the current study’s CI cohort received their implants, as the inclusion criteria specified only that this occurred before the age of 5 years. Early cochlear implantation has been linked with better developmental outcomes for children with aided hearing loss (Fulcher et al., 2012; Johnson & Goswami, 2010), so different results may have been obtained had all of the present study’s participants received their CIs at a younger age. While the reported results do still pertain to current clinical caseloads of children with CIs, future research may focus on children who receive CIs earlier in life (i.e., before the age of 12 months), in order to both monitor the outcomes of this cohort and to represent evolving clinical standards within Australia.

The large range of measures used in the present study allowed for an in-depth examination of many different skill areas. One limitation, however, was that the order of assessments was generally fixed across participants, which may have meant assessments administered at the end of test sessions were affected by examinee fatigue. That said, children with and without CIs would have been similarly affected by fatigue, such that fatigue should not have contributed significantly to the observed group differences.

2.5.2. Text-level reading profiles in beginning readers with cochlear implants. According to the Simple View of Reading model, reading comprehension is the product of two broad skillsets: word recognition and listening comprehension (Catts et al., 2005; 2015; Hoover & Gough, 1990). In the current study, this theoretical model was used as a framework within which the skills underlying reading comprehension were compared between groups. It was hypothesised that reading comprehension profiles would appear similar across TH and CI children, and indeed this hypothesis was largely supported. For both groups, reading comprehension was significantly dependent on listening comprehension and word reading skills. The profiles did, however, differ in terms of which predictor variable contributed the most variance to the regression model. The predominant variable for children with CIs was word reading accuracy, whereas for TH children it was listening comprehension. The TH group's pattern of performance may therefore reflect a higher degree of word recognition automaticity, which thus enables them to devote more cognitive resources to comprehending the text meaning (Language & Reading Consortium, 2015).

The present findings expand on past studies involving children with CIs that have found a similarly strong relationship between reading comprehension and single word reading skill (Geers, 2003; Johnson & Goswami, 2010; Vermeulen et al., 2007; von Muenster & Baker, 2014). One strength of the present study was that reading comprehension was analysed as the product of word reading and language comprehension, so the contribution made by each of these underlying skills was viewed in the context of the other. In two previous studies that have also based analyses on a Simple View of Reading framework, reading comprehension for children with CIs was significantly related to word recognition, but was equally or more strongly related to oral language (Vermeulen et al., 2007; von Muenster & Baker, 2014). Importantly though, the age of these CI cohorts ranged from 7 to 22 years old (Vermeulen et al., 2007) and 5 to 12 years old (von Muenster & Baker, 2014). In typically developing children, the skills underlying reading comprehension shift, so that language comprehension plays an increasingly important role as word recognition becomes more automatic (Catts et al., 2005; 2015; Garcia & Cain, 2014; Language & Reading Consortium, 2015). Hence, the relatively reduced dependence on oral language in the CI group of the present study may be attributed to the narrow age range of our participants, all of whom were in the first three years of formal literacy instruction.

Further to the above point, the CI group relied more strongly on word reading skills than even their TH peers to achieve reading comprehension. This finding provides unique insight into how reading comprehension skills in children with CIs compare to those with typical hearing. The observed pattern of results may reflect the CI group's delayed pattern of reading development, again given that younger typically developing readers tend to rely more on word recognition than older, more skilled readers (Catts et al., 2005; 2015; Garcia & Cain, 2014; Language & Reading Consortium, 2015). This interpretation of the findings does not discount the value of linguistic skill in achieving reading comprehension, since readers require a minimum level of receptive language competence to comprehend print once written words have been decoded. Indeed, reading comprehension appeared, on the whole, preserved for children with CIs, given that their average standard score was 98.43 and the group difference in scores only approached statistical significance. Still, word-level limitations should be expected to result in a comparatively reduced capacity to draw meaning from the text (Garcia & Cain, 2014), and it is possible that the CI group might have obtained even better outcomes had their word reading accuracy been as high as their TH peers. Future research may seek to determine whether the timing of implantation (i.e., early versus late) impacts the differential capacity of word recognition and listening comprehension to predict reading comprehension in early readers.

2.5.3. Word-level reading profiles in beginning readers with cochlear implants. There has been a substantial amount of research demonstrating the value of phonological awareness in predicting word reading achievement for beginning readers with CIs (Cupples, Ching, Crowe, Day & Seeto, 2013; James et al., 2009; Nittrouer et al., 2012). In the present study, phonological awareness was hypothesised to contribute significantly to word reading ability for both the CI and TH groups. This proved to be the case, thereby providing support for the assertion that skills in attending to the sublexical constituents of a word are critical for early reading development, regardless of hearing status (Mayer & Trezek, 2014).

Within the theorised architecture presumed to underlie single word recognition in skilled readers, phonological and orthographic processors both play important roles (Coltheart et al., 2001; Seidenberg & McClelland, 1989). As stated, orthographic information pertains to the letters in a given word, the stored representation of which – if familiar – can be partially or wholly retrieved when encountered in text (Hagiliassis et al., 2006). Previous research has focused much more on establishing the role of phonological processing in facilitating reading development for children with CIs, and relatively little focus has been directed towards orthographic processing. Some researchers have, however, posited that individuals with CIs 'compensate' for phonological processing limitations to the extent that they can use orthographic or lexical knowledge to successfully recognise words (Geers & Hayes, 2010; Nittrouer et al., 2014). It was on this basis that in the present study, the CI group's non-phonological skills were expected to contribute more so to word reading accuracy, relative to the TH group. That hypothesis was not supported by the findings. Although orthographic skill significantly predicted the majority of word reading variance for the CI group, this value was only slightly higher than that of the TH group. Hence, orthographic processing was a key component underlying single word recognition for all participants.

Despite drawing on underlying phonological and orthographic skills to a similar degree as TH children, the CI group still had significantly poorer overall word reading skills.

Future research in this area may therefore benefit from the inclusion of both chronological and reading age-matched peers to investigate the contribution of non-phonological skills to word reading. It is possible that the current study's CI group reached their level of word reading proficiency by relying more so on orthographic processing than what would have been observed in a (presumably younger) reading-matched TH group. Whether such a developmental trajectory can lead to successful reading outcomes that are commensurate with TH peers is currently unclear. Additional studies that directly test the effectiveness of specific interventions will be critical in informing clinicians and educators as to the most appropriate treatment approach for facilitating literacy improvements in this population.

Semantic processing, which was represented in the current study by receptive vocabulary performance, was also included as an expected predictor of word-level reading accuracy. Isolated from other measures, it did significantly relate to word reading performance. However, it failed to contribute to the regression model when entered simultaneously with phonological awareness and orthographic processing. This pattern of results pointed to a high degree of collinearity between vocabulary and other word-level skills, which thus makes it difficult to determine the true significance of its independent contribution to word reading. Additionally, since the outcome variable in these analyses represented *single* word recognition, items presented in this assessment were isolated from context. Hence, semantic skills would certainly be expected to play a greater role in facilitating recognition of words contextualised in a written passage.

Past studies have shown that semantic processing plays an important role in literacy development. In the context of the hearing-impaired population, James and colleagues (2009) found that word reading accuracy for school-age children with CIs was significantly and predominantly predicted by receptive vocabulary. In the same study, phonological (i.e., rhyme) awareness also contributed significant unique variance to word reading in this group, but only when vocabulary was not accounted for. As was the case in the present study, collinearity between predictor variables may therefore have obscured the exact unique value of phonological and semantic processing. Without contradicting the position that vocabulary is critical to early reading development, the current study findings further highlight the interconnected nature of semantic and other underlying word-level skills.

2.6. Conclusion

This study investigated the early reading development of children with CIs who all used spoken language only and received early auditory-verbal intervention. Our findings showed that these children had significantly worse spoken language and word reading accuracy than their peers with typical hearing. However, their reading comprehension was relatively preserved, which may reflect the present CI cohort's homogeneity and early exposure to spoken language-based intervention. Further group comparisons indicated that the CI group showed increased reliance on word reading, and this may represent a delay in the development of word processing automaticity. Word reading, for children with CIs and their TH peers, was found to depend on both orthographic and phonological skills. As a relatively unexplored area of research, results from the present study pertaining to word- and text-level reading profiles provide the impetus for future research to explore how variables (such as age at implantation) impact outcomes. Additional insight may also be gained from longitudinal investigations that track the reading outcomes and profiles of beginning readers with CIs over time.

Chapter 3.

Spelling Development in Children with Cochlear Implants: Evidence of Underlying Processing Differences.

In Chapter 2, the reading outcomes of children with cochlear implants were explored, with reference to existing theoretical models of word- and text-level written language processing. In Chapter 3, a similar psycholinguistic approach will be taken to examine the *spelling* outcomes of the same two cohorts of children with cochlear implants and children with typical hearing. While Chapters 2 and 3 provide separate insights into the thesis topic, together they also contribute to a comprehensive understanding about literacy development in children with cochlear implants, as measured behaviourally.

Chapter 3 presents a corrected version of an article submitted for journal publication[1], which is currently under revision. Minor changes have been made to formatting and methodological description, in order to maintain thesis continuity. A section on statistical power analyses has also been added to this chapter's 'Results' section. The authors' contributions to the original submitted article are detailed on the following page.

[1] Bell, N., Angwin, A.J., Wilson, W.J., & Arnott, W.L. (2018*). Spelling in children with cochlear implants: evidence of underlying processing differences.* Manuscript under revision.

Contributor	Statement of contribution
Nicola Bell (Candidate)	Conception and design: 75% Data collection: 100% Data analysis: 90% Interpretation: 80% Manuscript writing: 100% Manuscript revisions: 20%
Dr Anthony Angwin	Conception and design: 5% Interpretation: 5% Manuscript revisions: 30%
A/Prof Wayne Wilson	Conception and design: 5% Interpretation: 5% Manuscript revisions: 20%
Dr Wendy Arnott	Conception and design: 15% Data analysis: 10% Interpretation: 10% Manuscript revisions: 30%

3.1. Abstract

This study compared the spelling skills and sub-skills of young children with cochlear implants (CIs) who use spoken language only (n = 14) with those of a same-aged typically hearing (TH) control group (n = 30). Spelling accuracy was assessed using irregular and nonsense word stimuli. Error and regression analyses were conducted to provide insight into the phonological and orthographic spelling strategies used by each group. Results indicated that children with CIs were as accurate as the TH group. However, misspellings made by the CI group were less phonologically plausible, and while nonword spelling accuracy was related to letter-sound knowledge for the TH group, the same relationship was non-significant for the CI group. Hence, despite demonstrating a similar degree of overall spelling success to TH children, children with CIs appeared to apply phonics skills less effectively.

3.2. Introduction

On average, children with severe-to-profound hearing loss demonstrate literacy skills that are below the level of their typically hearing peers (Harris, 2015). For all beginning readers, learning to decode words relies on their knowledge of how letters correspond with speech sounds (McBride-Chang, 1999). As such, the ability to process phonological information is key to facilitating successful literacy development, and indeed deficits in such skills are often presumed to underlie disordered reading in children with and without hearing loss (Mayer & Trezek, 2014). While there are abundant studies investigating the reading development of individuals with hearing loss, less is known about their spelling. This reflects a clinically significant gap in evidence, since to be 'literate', one must be able to both decipher and produce written text. The present study examined the spelling skills, and the underlying processes contributing to those skills, in young children with cochlear implants (CIs) who use spoken language to communicate.

3.2.1. Phonological processing in children with hearing loss. 'Phonological processing' is a term used to describe the explicit or implicit computation of spoken word constituents (Wagner, Torgesen & Rashotte, 1994). Belonging to this broad category of literacy sub-skills are *phonological awareness*, (i.e., the identification and/or manipulation of spoken word parts) and *phonological memory* (i.e., the short-term storage and retrieval of phonological information; Wagner et al., 1994). Phonological processing skills also pertain to an individual's knowledge and application of the relationships between speech sounds and letters (Loveall, Channell, Phillips & Conners, 2013), and such *phonics* abilities are key to early reading and spelling development (Castles, Rastle & Nation, 2018). In the present study, children were assessed on their phonological processing skills, using measures of phonological awareness, phonological memory and letter-sound knowledge. Previous research has found that children with significant hearing loss generally score lower on such tasks than their typically hearing peers (Ambrose, Fey & Eisenberg, 2012; James, Rajput, Brinton & Goswami, 2008; Nittrouer, Caldwell, Lowenstein, Tarr & Holloman, 2012).

Some researchers have posited that individuals with hearing loss 'compensate' for phonological processing limitations by relying on non-phonological skills, such as orthographic and semantic processing, to perform literacy tasks (Geers & Hayes, 2010; Kyle & Harris, 2006; Nittrouer, Caldwell-Tarr, Sansom, Twersky & Lowenstein, 2014). Furthermore, children with significant hearing loss may draw differentially on underlying skills, depending on whether they are reading or spelling. According to a study by Kyle and

Harris (2011), the reading abilities of 7- and 8-year-old children with aided severe-to-profound hearing loss were predicted by speechreading, vocabulary and letter-sound knowledge skills, as measured two years prior. The group's spelling abilities, however, were only predicted by earlier letter name knowledge. Hence, the reading and spelling profiles of these children appeared qualitatively distinct. In support of a theorised disparity between reading and spelling developmental trajectories for children with severe-to-profound hearing loss, there appears to be a trend in the results reported in the literature whereby this population demonstrates below-average reading abilities, but age-appropriate spelling abilities (, Keetay, Boyd, Palmatier & Wacks, 1998; Burden & Campbell, 1994; Colin, Leybaert, Ecalle & Magnan, 2013; Harris & Moreno, 2004).

3.2.2. Spelling outcomes in children with hearing loss. In order to investigate what spelling strategies are employed by children with hearing loss, a number of previous studies have conducted spelling error analyses. For example, Roy, Shergold, Kyle & Herman (2015) examined the phonological plausibility of spelling errors produced by 11-year-old children with aided severe-to-profound hearing loss. Items categorised as 'plausible' retained all or most of the misspelled word's phonological structure (e.g., 'allthough' for 'although'). 'Implausible' errors were more significantly misspelled, so pertained to items where multiple *phonemes* (i.e., speech sounds) were omitted, substituted or incorrectly ordered. As a group, the proportion of phonologically plausible errors made by children with hearing loss was 52.7%, which was significantly lower than the proportion produced by a comparison group of typically hearing children with dyslexia (79.81%). Children with hearing loss therefore appeared to rely less on a method of converting the phonological constituents of target words to corresponding letter sequences than their hearing counterparts (Roy et al., 2015). Interestingly, despite the discrepancy, overall spelling accuracy for the group with hearing loss was still similar to the group with dyslexia, indicating that the same degree of success was obtained via different strategies. Other studies involving spelling error analyses have also found that the proportion of phonologically plausible errors is reduced in children with severe-to-profound hearing loss, when compared with same-aged typically hearing children of average reading ability (Harris & Moreno, 2004; Hayes, Kessler & Treiman, 2011).

3.2.2.1. Spelling outcomes in children with cochlear implants. A high percentage of children with significant hearing loss receive cochlear implants (CIs), and can effectively communicate using spoken language by way of this technology. However, while capturing auditory stimuli well enough for users to orally communicate, CIs do not provide the same degree of spectral clarity for speech as is provided by a typically functioning cochlea (Nittrouer et al., 2012). Phonological processing difficulties are therefore still commonly observed in children with CIs (Ambrose et al., 2012; Fitzpatrick et al., 2012; Lee, Yim & Sim, 2012; Nittrouer et al., 2012), and as with hearing children, such limitations are thought to play a causal role in concomitant literacy difficulties (Mayer & Trezek, 2014; Nittrouer et al., 2012).

There is evidence to suggest that both young children and adolescents with CIs experience difficulties with spelling (Apel & Masterson, 2015; Geers & Hayes, 2010). Apel and Masterson (2015) analysed the written word productions of nine children with CIs, whose mean age was 8;11 years. The group obtained a lower overall spelling accuracy score than a typically hearing group of children at the same reading level, although this difference did not reach statistical significance. Given that the study's reading-matched control group was almost a year younger than the CI group, it may be assumed that the children with CIs would have scored lower than typically hearing children of the same age. However, standardised scores were not reported, so the exact degree of divergence from age-based norms is unclear. In a similar study, Geers and Hayes (2010) investigated the outcomes of older adolescents aged 15 to 18 years. Significantly more errors were produced by the CI group, compared to a typically hearing group matched on chronological age. While these findings, as well as those reported by Apel and Masterson (2015), suggest that children with CIs demonstrate more spelling difficulties relative to their peers, it may be noted that both studies' cohorts consisted of individuals from varied linguistic backgrounds. Given that communication mode has been found to influence literacy outcomes in children with hearing loss (Cupples et al., 2017; Johnson & Goswami, 2010; O'Donoghue, Nikopoulos & Archbold, 2000), the skills of spoken language users may be better represented when analysed separately from sign language users.

A number of studies have provided evidence as to the spelling skills of spoken communicators with CIs (Fitzpatrick et al., 2012; Hayes et al., 2011; Straley, Werfel & Hendricks, 2016). Fitzpatrick and colleagues (2012) administered a standardised real word spelling-to-dictation task to children with significant hearing loss aged 6 to 18 years, who used spoken language and received CIs at 2.9 years (SD = 1.2y). Compared with the test's normative sample (whose mean score was 100), the CI cohort achieved an average standard score of 103.7. Moreover, 80% of participants' scores were within normal limits. Similarly, Hayes et al. (2011) examined the spelling abilities of oral communicators aged 6 to 12 years, who received CIs at 3 years (SD = 1.3y). Using an experimental assessment task, wherein participants were asked to name, and then spell, a pictured item, the authors found no significant difference in performance between the CI cohort and a similarly aged group of typically hearing children. Importantly however, since all spelling analyses in the study by Hayes et al. (2011) were conducted whilst controlling for reading comprehension scores, it is possible that spelling accuracy for children with CIs deviated significantly from the age-matched cohort's, but that reading comprehension scores also deviated to the same degree. Group differences in reading comprehension and standardised scores on reading and spelling measures were not reported, so this possibility cannot be confirmed.

The above results for children with CIs who are oral communicators, while promising, contrast somewhat with those reported by Straley et al. (2016). Children in Straley and colleagues' (2016) study, who were aged between 8 and 12 years, achieved a mean standardised spelling score of 92.6, which is still within normal limits, but more than ten standard score points below the cohort examined by Fitzpatrick and colleagues (2012). It is not clear what accounts for the difference in reported outcomes, since the spelling assessment measures appear similar across studies, as do the hearing technologies, linguistic backgrounds, ages and nonverbal intelligence scores of participants. Straley and colleagues (2016) did not report the age at which participants received their CIs, although the age at which 'amplification' was provided – presumably via hearing aids – was slightly older and more variable (mean = 1.6y; SD = 1.8y) than participants in the Fitzpatrick et al. (2012) study (mean = 1.3y; SD = 0.6y). Since early amplification and implantation are associated with better spoken and written language outcomes (Johnson & Goswami, 2010), this discrepancy may account for some of the difference. Importantly though, no typically hearing control group was included in the Fitzpatrick et al. (2012) and the Straley et al. (2016) studies, which limits the degree to which the CI groups' spelling skills might be directly compared with each other or with overall age-based expectations.

Presently, there is very little research concerning the spelling strategies used by oral communicators with CIs. In the aforementioned study by Hayes and colleagues (2011), this gap was addressed with reference to analyses of the children's spelling errors. The authors found that children with CIs produced fewer phonologically plausible errors than typically hearing children, which is consistent with previous observations in the wider population of children with hearing loss. Within the same study, different word characteristics were examined to determine how spelling accuracy was differentially influenced between the two groups. One of the characteristics was word ‘typicality’, which was defined as the consistency with which a letter or letter sequence represented – in general English vocabulary – a certain phoneme. The hearing group was more accurate when producing words with typical spellings (e.g., ‘stamp’), compared with words that had atypical spellings (e.g., ‘scissors’). Children with CIs also demonstrated this effect, though to a significantly lesser extent. The authors attributed the CI group’s diminished typicality effect to reduced phonological sensitivity, since atypical words rely on visual memorisation strategies more than phonological knowledge (Hayes et al., 2011).

Importantly, regardless of regularity, a speller is able to produce a familiar real word using visual memory strategies (Houghton & Zorzi, 2003). Nonwords, on the other hand, cannot be retrieved from memory as existing lexical representations. Rather, individuals must determine for themselves how each phoneme in the item is best represented by one or multiple letters. Accordingly, nonword spelling provides a more reliable measure of a person’s ability to draw on their phonological knowledge than regular (or ‘typical’) real word spelling. To the author’s knowledge, only one study has examined nonword spelling in children with hearing loss. Nelson and Crumpton (2015) reported that children with various hearing loss severities and technology configurations had significant difficulties with this task, when compared with age-matched typically hearing peers. Interestingly too, phonemic awareness was found to predict concurrent nonword spelling accuracy for typically hearing children, but not for children with hearing loss. Given the scarcity of evidence regarding nonword spelling, further research is needed to examine its relationship with phonological processing in children with CIs. To this end, nonword spelling was examined in the present study, with respect to the skills demonstrated by children with bilateral severe-to-profound hearing loss, who use spoken communication and have CIs. Unlike the cohort in Nelson and Crumpton (2015), the cohort included here also received auditory-verbal therapy (AVT) from an early age.

Auditory-verbal therapy is an intervention approach for individuals with hearing loss, which aims to facilitate language development through solely spoken communication. The approach’s emphases on speech and oral language have been theorised to promote emergent literacy skills, by drawing explicit attention to words’ phonological structures (Kaderavek & Pakulski, 2007). Yet, of the studies investigating written language proficiency in children who have undergone AVT (e.g., Dornan, Hickson, Murdoch, Houston & Constantinescu, 2010; von Muenster & Baker, 2014), none has compared results with a typically hearing age-matched control group. In addition, prior research involving AVT-exposed children has focused on spoken language or reading outcomes, rather than spelling. The present study aims to address this gap in evidence, by investigating the spelling performances and profiles of children with CIs who use spoken communication and have received AVT.

3.2.3. Current study. Broadly, children with severe-to-profound hearing loss show spelling difficulties, relative to typically hearing children of the same chronological age (Arfé, Rossi & Sicoli, 2015; Harris & Moreno, 2004; Roy et al., 2015), although they may perform similarly to or better than those who are matched on reading ability (Aaron et al., 1998; Burden & Campbell, 1994; Colin et al., 2013; Harris & Moreno, 2004). Within the subset of children with hearing loss and CIs, it is not clear how well spelling accuracy aligns with age-based expectations, since most studies have compared the group's results with either other children with hearing loss (Fitzpatrick et al., 2012; Harris & Terlektsi, 2011) or typically hearing children whose reading abilities are matched or controlled (Apel & Masterson, 2015; Hayes et al., 2011). Additionally, only a few studies have examined children with CIs who use spoken language only to communicate, despite the finding that linguistic mode may influence outcomes (Cupples et al., 2017).

Clearly, further research is needed to investigate spelling skills in children who are homogeneous not only in terms of age and hearing device, but also in terms of their mode of communication. To that end, the first research question addressed in the current study was: How do the spelling skills of young children with CIs, who use spoken language, compare with those of typically hearing children of the same age? It was expected, based on results reported for other similarly aged cohorts (e.g., Apel & Masterson, 2015), that children with CIs would demonstrate poorer spelling accuracy than children with typical hearing. However, the question was treated as exploratory, given the scarcity of existing research in which irregular and nonsense word spelling accuracy have been examined separately.

The present study also examined the contribution of underlying skills to spelling outcomes in young CI users. The second research question was: Do underlying linguistic processing skills contribute to spelling outcomes differently in young children with CIs, who use spoken language, compared with typically hearing children of the same age? Based on past research findings reported by Hayes and colleagues (2011), it was expected that the CI group would produce a disproportionately high number of spelling errors that were phonologically implausible. The second research question was also informed by regression analyses, which were included to examine the relationships between children's underlying phonological and orthographic skills and their spelling accuracy outcomes. This latter approach was considered exploratory in nature, again given the lack of existing research with separate irregular and nonsense word spelling outcome measures; hence, no hypothesis was formulated for the regression analyses.

3.3. Method

3.3.1. Participants. Fourteen children with CIs (7 females, 3 left-handed) and 31 typically hearing (TH) children (16 females, 3 left-handed) participated in the present study. Most children with CIs and many of the TH participants were recruited from Hear and Say, an organisation that provides audiological and spoken language intervention to children with hearing loss, as well as conducting regular school-based hearing screening. Parents and caregivers who had given permission to be contacted for research purposes were invited to participate in the study. The remaining participants were recruited from the wider community via newsletter advertisements and word of mouth.

All participants adhered to the following inclusion criteria: (1) use of spoken English as native and primary form of communication; (2) nonverbal reasoning at or above normal limits (as measured using the *Raven's Coloured Progressive Matrices*; Cotton et al., 2005; Raven, Raven & Court, 2004); (3) no diagnosed developmental disorders or intellectual disabilities, and; (4) in Grade 1, 2 or 3 of a mainstream school. The CI and TH groups did not differ on gender, $\chi^2(1) = 0.010$, $p = 0.920$, handedness, $\chi^2(1) = 1.153$, $p = 0.283$, age, $t(43) = -1.483$, $p = 0.145$, grade, $\chi^2(1) = 1.539$, $p = 0.463$, or nonverbal reasoning, $t(43) = 0.664$, $p = 0.510$. With regard to nonverbal reasoning, 'normal limits' was operationally defined as performance within one standard deviation of the mean (i.e., a z-score on the *Raven's Coloured Progressive Matrices* of between -1 and +1). Age and nonverbal reasoning scores were checked to confirm that data were normally distributed within each group and variances were equal between groups. There were no outliers for age or nonverbal reasoning in either group.

Additional inclusion criteria for the children with CIs were also implemented: participants needed to have received their CIs before the age of 5 years, and have received AVT before entering formal schooling. In all cases, AVT was administered by AVT-certified speech pathologists or AVT-certified Teachers of the Deaf. The mean age of the CI group at the time of testing was 8.01 years (SD = 0.82y). These participants were in Grade 1 (n = 1), Grade 2 (n = 6) or Grade 3 (n = 7). The CI group's average nonverbal reasoning z-score on the *Raven's Coloured Progressive Matrices* was 0.72 (SD = 1.21). All children received CI surgery before the age of 5 years (mean = 1.74y; SD = 1.45y), and the mean duration of CI use at the first point of current testing was 6.23 years (SD = 1.52y). Information pertaining to each participant is shown in Table 3.1 (see Chapter 2 of this thesis for additional details relating to the CI cohort).

Participants in the TH control group passed hearing screening assessments (thresholds of 25dB HL or better at octave intervals of 500 to 4000 Hz) conducted using a commercially available screening audiometer. The participants completed these screenings after entering formal schooling and no more than two years prior to their participation in the present study. The mean age of the TH group was 7.63 years (SD = 0.77y). Five participants were in Grade 1, 16 were in Grade 2, and 10 were in Grade 3 at the time of testing. The group's average nonverbal reasoning z-score on the *Raven's Coloured Progressive Matrices* was 0.95 (SD = 0.98).

3.3.2. Ethics statement. Ethical approval for the present study was obtained from the Behavioural and Social Sciences Ethical Review Committee at the University of Queensland. Gatekeeper ethical approval was also obtained from the Hear and Say Research and Ethical Advisory Committee. All parents gave written informed consent for their children to participate in the study.

Table 3.1

Audiometric information for participants with CIs (n=14).

#	Configuration	Aetiology	Stable (Y/N)	Age (m) at 1st implant	Age (m) at EI enrolment	Unaided PTA (dB)		Aided PTA (dB)	
						Left	Right	Left	Right
1	CI+CI	Con. (idiopathic)	Y	46	2	75.00	76.25	25.00	25.00
2	CI+CI	Con. (idiopathic)	Y	7	1	98.75	≥100	26.25	25.00
3	CI+CI	Con. (idiopathic)	Y	7	2	92.50	92.50	25.00	26.25
4	CI+CI	Con. (idiopathic)	N	37	29	75.00	63.75	22.50	22.50
5	CI+CI	Con. (idiopathic)	N	31	3	88.75	82.50	22.50	18.75
6	CI+CI	Con. (idiopathic; LVAS)	Y	9	4	76.25	97.50	21.25	22.50
7	CI+CI	Con. (genetic-NS)	Y	8	2	98.75	98.75	25.00	22.50
8	CI+CI	Con. (genetic-Connexin 26)	Y	7	1	≥100	≥100	22.50	18.75
9	CI+CI	Con. (genetic-Connexin 26)	Y	8	1	95.00	≥100	22.50	21.25
10	CI+CI	Acq. (1;3y; idiopathic)	Y	19	31	≥100	≥100	27.50	22.50
11	CI+CI	Con. (CMV)	Y	8	4	≥100	98.75	25.00	21.25
12	CI+CI	Con. (CMV)	Y	8	7	97.50	≥100	30.00	28.75
13	CI+HA	Con. (genetic-Pendred Syndrome)	N	47	1	96.25	75.00	28.75	*
14	HA+CI	Acq. (1;3y; idiopathic; LVAS)	N	50	39	61.25	77.50	30.00	30.00

Note. CI = cochlear implant; HA = hearing aid; EI = early intervention; PTA = pure tone average; Con. = congenital; Acq. = acquired; LVAS = Large Vestibular Aqueduct Syndrome; NS = not specified; CMV = Cytomegalovirus. *Data for left ear missing but reportedly similar to right ear.

3.3.3. Tests and materials.

3.3.3.1. Spelling. Written single word spelling was assessed using the *Diagnostic Spelling Test – Irregular Words* (DiSTi; Kohnen, Colenbrander & Nickels, 2012) and the *Diagnostic Spelling Test – Nonwords* (DiSTn; Kohnen, Colenbrander, Krajenbrink & Nickels, 2013), both of which are normed on Australian children in Grades 1 through 7. There are 74 items on each test, and in accordance with the test guidelines, both assessments are discontinued if the examinee reaches a ceiling of five incorrect responses. Responses are scored '0' if inaccurate and '1' if accurate. The test items on each test were dictated twice (or more, if requested by the child). For the *DiSTi*, each item was also produced in the context of a sentence, as per the test protocol. The test administrator articulated each item clearly, but did not draw attention towards, or away from, her mouth while dictating the spelling targets. Participants from either group may have received additional visual cues concerning the articulatory characteristics of target items (and in particular, of nonwords), but this was not actively manipulated during testing.

In the *DiSTi*, examinees are asked to write the irregular word that is dictated to them by the test administrator. Results from the *DiSTi* provide a good indication of how well examinees spell real words that cannot otherwise be coded using letter-sound correspondence knowledge. At the beginning of the *DiSTi*, stimulus words are frequent and short (e.g., item 1: 'good'). As the list progresses, stimuli comprise more complex and infrequent words (e.g., item 74: 'mayonnaise').

In the *DiSTn*, examinees are asked to write the nonword that is dictated to them by the test administrator. Since the stimuli in the *DiSTn* are nonwords, they cannot be produced through any other process than by applying knowledge of letter-sound correspondences. The rationale for including such a measure is that participants must rely on their phonics knowledge to produce items, rather than their knowledge of familiar orthographic representations. The *DiSTn* therefore allows for children's phonics knowledge, as it relates to spelling performance, to be captured. Responses are scored as accurate if they plausibly represent the pronunciation of the target word (e.g., acceptable responses for item 14: 'leat', 'lete' or 'leet'). Stimuli are all one-syllable in length, but contain increasingly uncommon vowel spelling patterns and complex consonant-vowel combinations (e.g., item 1: 'mip', item 74: 'zoish').

3.3.3.2. Spelling Sensitivity Score (SSS). Responses to the *DiSTi* were examined to determine each participant's Spelling Sensitivity Scores for both elements (SSS-E) and words (SSS-W). The protocol for scoring was originally described by Masterson and Apel (2010), and has been used previously with CI cohorts (e.g., Apel & Masterson, 2015; Straley et al., 2016). By definition, items in the *DiSTi* contain letter sequences that do not commonly represent English phonemes and must therefore be learned as whole or partial orthographic representations. The errors produced in response to such items provided insight into the underlying skills contributing to each individual's spelling outcomes. Responses were scored along a gradient, with phonologically plausible elements receiving a higher score than those that were implausible. For this reason, the same analysis could not be performed for the *DiSTn*, since any response that plausibly approximated the nonword item's pronunciation was categorically correct, with no middle ground between this outcome and what was categorically incorrect. The SSS analyses were therefore only conducted with *DiSTi* errors.

In prior studies where the SSS has been used to analyse spelling, all examinees were reported to have produced the same or a similar number of total words, and analyses included all responses – both correct and incorrect (e.g., Apel & Masterson, 2015). In the present study however, the total number of words varied significantly between participants, depending on the item at which ceiling performance on the *DiSTi* was reached. For this reason, SSS analyses were conducted on incorrect responses only. In addition, the aim of implementing the SSS was to examine the spelling strategies used by participants, based on the types of errors they made, whereas the difference in overall accuracy between groups was examined separately with reference to standardised *DiSTi* and *DiSTn* scores. As described in further detail below, each participant's SSS-E and SSS-W values were averaged across their total number of misspellings, thereby accounting for different numbers of errors made between participants.

Spellings were scored by the first author. A second independent scorer checked element scores for 50% of the total participants (i.e., half from each group). Inter-rater reliability, as indexed by the intra-class coefficient (ICC) value, was calculated using a mean-rating, two-way mixed model. The ICC for absolute accuracy and consistency was 99.3%, which represents excellent reliability (Koo & Li, 2016). The rare examples of disagreement were resolved by discussion between the two main scorers and a third independent scorer. All three spelling scorers were qualified speech pathologists. Following discussions with the second and third scorers, the first author re-analysed the remaining 50% of participants, in order to ensure consistency.

3.3.3.2.1. Spelling Sensitivity Score – Elements (SSS-E). Based on the protocol outlined by Masterson and Apel (2010), incorrect responses elicited in the *DiSTi* were first segmented into 'elements'. These elements were defined as the individual phonemes in a base word (e.g., g/oo/d), or for multi-morphemic words, the affix and the juncture change (e.g., s/t/o/p/p/ing). Depending on linguistic accuracy and plausibility, each element was then scored on a 4-point scale. A score of '3' was assigned to correctly spelled elements. A score of '2' was assigned to elements which were orthographically legal and phonemically plausible (e.g., 'skool' or 'fortunet'). Such errors were presumed to reflect the participant's awareness of the target phoneme, and their knowledge about how it may commonly be spelled in other contexts. An element whose presence was marked with an orthographically or phonologically illegal letter sequence received a score of '1' (e.g., 'spool' or 'schoool' for 'school'). These errors may be attributed to reduced awareness of how phonemes and morphemes are represented by letters (Apel, 2011). Finally, where the target element was omitted, participants received a score of '0' (e.g., 's[ ]ool' for 'school'). Unlike plausible and implausible misspellings, element omissions were seen to represent reduced awareness of the word's basic phonemic structure (Apel & Masterson, 2015).

After scoring elements according to the above protocol, each item's SSS-E was calculated by dividing the sum of element scores by the total potential element score in the word. An average SSS-E was then calculated for each participant and used for all subsequent analyses. Again with reference to the scoring procedures described by Apel and Masterson (2015), incorrectly spelled items were also subjected to an examination of error types at a whole word level, as described below.

3.3.3.2.2. Spelling Sensitivity Score – Words (SSS-W). The SSS-W was calculated to represent the types of errors observed in words as a whole, rather than at the element level. This form of analysis is reminiscent of many previous studies, in which whole words are categorised as having phonologically plausible or implausible spellings (e.g., Harris & Moreno, 2004; Hayes et al., 2011). Words containing only orthographically and phonologically legal spellings (i.e., homophones or pseudohomophones) scored '2'. Phonologically *im*plausible spelling errors, in which words contained elements represented by phonologically or orthographically illegal letter sequences, scored a SSS-W of '1'. Words where elements had been omitted scored '0'.

Each participant received a SSS-W score, which was calculated as the average of their individual item scores. Given the emphasis in past studies on phonological plausibility of spelling errors, the frequencies with which participants produced scores of ‘2’ were also examined for group differences. Additional follow-up analyses explored the frequencies with which the other two implausible error types (i.e., those that received scores of ‘0’ or ‘1’) were produced by CI and TH groups.

3.3.3.3. Spelling sub-skills. A number of underlying skills were presumed to contribute to participants’ overall spelling outcomes. Those selected for analysis in the present study pertained to phonological processing (i.e., phonological awareness, phonological memory, letter-sound knowledge) and orthographic processing. The rationale for choosing these particular measures was based on evidence that phonological and orthographic processing skills contribute significantly to literacy development (Houghton & Zorzi, 2003). Whether children with CIs compensate for phonological processing difficulties by relying more so on orthographic processing skills (as has been posited previously; e.g., Hayes et al., 2011) was also a key question addressed in the present study. The measures themselves and the procedures for administering them are summarised below.

3.3.3.3.1. Phonological awareness. Three subtests from the *Comprehensive Test of Phonological Processing – 2nd edition* (*CTOPP-2*; Wagner, Torgesen, Rashotte & Pearson, 2013) were administered to assess phonological awareness. The CTOPP-2 is a norm-referenced test, with standardised scores available for children aged 4 and over. An index score representing phonological awareness skill was calculated on the basis of children’s performance on: (1) *Elision*; (2) *Blending Words*; (3) *Sound Matching* (4-7 years), and; (4) *Phoneme Isolation* (7+ years). In subtest (1), examinees were asked to produce a given word with one phoneme omitted (e.g., ‘What is “flame” without /f/?”). In subtest (2), examinees were given isolated spoken word parts and asked to blend them together to form a word (e.g., ‘What is /s/ - /t/ - /æ/ - /m/ - /p/?’. In subtest (3), examinees (4-7y) were asked to match a pictured target word with one of three or four other pictured words, based on a common phoneme (e.g., ‘Which of these words has the same first sound as “cat”: “rug”, “car”, or “mat”?’). In subtest (4), examinees (7y+) were asked to identify the target phoneme from a spoken word (e.g., ‘What is the third sound in “frog”?’). As outlined in the test manual, raw scores from each of these subtests were combined and converted to a composite phonological awareness score.

3.3.3.3.2. Phonological memory. Phonological memory was assessed using the *Children's Test of Nonword Repetition* (*CNRep*; Gathercole & Baddeley, 1996). Standardised scores and percentile ranks are available for children aged 4;0 to 9;11 years. In this test, children are asked to repeat a given nonword. There are 40 items on the test, divided equally into two-, three-, four- and five-syllable nonwords. Nonword repetition has been found to reflect phonological processing skills, and is a significant predictor of reading ability in children (Wagner et al., 1994).

3.3.3.3.3. Letter-sound knowledge. Letter-sound knowledge is an important precursor to reading development (Catts, Herrera, Nielson & Bridges, 2015). The *Letter-Sound Test* (*LeST*; Larsen, Kohnen, McArthur & Nickels, 2011) was used to measure children's explicit knowledge of phoneme-grapheme correspondences. Australian norms for children aged 5;0 to 9;11 years were used to convert raw scores to standardised z-scores (Larsen, Kohnen, Nickels & McArthur, 2015). In this task, examinees are visually presented with 51 individual letters (or letter groups like 'ch') and are asked to say aloud the associated sound. Items are scored either '0' (incorrect) or '1' (correct), and the total raw score can be converted to a standardised score.

3.3.3.3.4. Orthographic processing. Orthographic processing was assessed using the *Test of Orthographic Choice* (*TOC*; Kohnen, Anandakumar, McArthur & Castles, 2012). This assessment, which has standardised norms for Grades 1 through 6, measures written word recognition. Examinees are presented with two words, both of which may be pronounced identically, but only one of which is a real word (e.g., 'caip'/'cape'). Children are asked to circle which of the items is a real word. The test contains 30 items and two practice items. Accuracy depends on retrieval of the word's orthographic representation, rather than knowledge of the word's meaning or the ability to 'sound it out'. Raw scores out of 30 were converted into standardised z-scores.

3.3.4. Procedure. The spelling measures reported above were administered as part of a larger language and literacy assessment battery (see Chapter 2). All participants were individually administered the assessments in a quiet room. The standardised protocols were adhered to, and all assessments were administered by the same qualified speech pathologist (author Bell). The spelling-to-dictation tasks (i.e., the *DiSTi* and *DiSTn*) were, as previously stated, administered on a face-to-face basis, with consistent pronunciations provided for all participants. The assessments were scored during the testing session where possible. The phonological awareness and memory tasks were aurally recorded using a Philips Voicetracer

620, so that any responses that were missed or unclear could be verified at a later time by the same test administrator.

3.4. Results

3.4.1. Spelling accuracy. The first research question pertained to the overall spelling scores of the CI and TH groups. These group comparison analyses were two-tailed, and standardised scores served as the dependent variables of interest. Upon inspecting the data distribution in each group, one outlier from the TH group was identified. This child's nonword spelling score (raw score = 0) was significantly lower than the rest of their cohort and so their nonword spelling data were subsequently excluded from analyses, due to concerns about the potential decrease in sample representativeness. Data were otherwise found to be distributed normally for each group, based on visual inspection of histogram plots and Levene's test for equality of variances.

Results from the group comparison are shown in Table 3.2. No statistically significant differences were found between the average irregular word spelling scores obtained by CI and TH groups. The difference between groups approached significance for nonword spelling (p = 0.064), with the CI group scoring lower than the TH group.

Table 3.2

CI vs. TH group comparisons for spelling and sub-skill measures.

Assessment measure	**CI (n=14) Mean (SD)**	**TH (n=31) Mean (SD)**	***t***	**Cohen's *d***	***p***
Irregular word spelling	.04 (1.25)	.33 (1.21)	.73	.23	.467
Nonword spelling[1]	-.05 (1.25)	.69 (1.19)	1.91	.61	.064
Phonological awareness[3]	90.71 (13.48)	107.45 (14.51)	3.66	1.19	*<.001
Phonological memory[3]	-1.33 (1.02)	0.28 (0.88)	5.44	1.70	<.001
Letter-sound knowledge[23]	-0.34 (1.13)	0.30 (0.97)	1.93	.61	.030
Orthographic processing	0.11 (1.25)	0.36 (1.22)	.62	.20	.541

Note. CI = cochlear implant; TH = typically hearing. Statistically significant values (p<.05) indicated with asterisk (*). Standardised z-scores reported for all measures (mean = 0.0) except Phonological Awareness, for which the Standard Score (mean = 100) is reported; [1]n=29 and [2]n=30 for TH group due to missing values for one participant and removal of one outlier in *DiSTn*; [3]p-value obtained using 1-tailed analyses.

3.4.2. Skills underlying spelling performance. The second research question focused on the skills underlying the above results. In order to investigate this question, the spelling error patterns produced by all individuals were also examined, as were the relationships between predictor skills and spelling outcomes. For all analyses, CI group results were compared against those of the TH group.

3.4.2.1. Spelling error analysis. One-tailed independent samples t-tests were conducted for the SSS-E and SSS-W values obtained by each group. Children with CIs obtained a mean SSS-E score of 2.37 (SD = 0.22), while the TH group obtained a mean score of 2.47 (SD = 0.13). The difference between groups was statistically significant, $t(43) = 1.82$, $p = 0.038$, and this was associated with a medium effect size. With respect to SSS-W scores, the CI group obtained a mean score of 1.24 (SD = 0.29), while the TH group obtained a mean score of 1.36 (SD = 0.23). This group difference in SSS-W scores approached statistical significance, $t(43) = 1.51$, $p = 0.070$.

Of the total number of inaccurate words produced by children with CIs, 36.11% contained phonologically and orthographically legal misspellings (i.e., received SSS-W scores of ‘2’). In contrast, the same type of errors made up 48.59% of the total inaccurate words for the TH group. The results of a one-tailed independent samples t-test confirmed that the difference between groups was significant, $t(43) = 1.752$, $p = 0.044$, and this corresponded with a medium effect size.

Follow-up analyses were conducted to explore potential group differences in the types of implausible errors produced by the groups. Phonologically or orthographically illegal letter substitutions (with SSS-W scores of ‘1’) comprised 50.69% of the CI group’s total errors, and 38.87% of the TH group’s total errors. Element omission errors (with SSS-W scores of ‘0’) comprised 13.19% of the CI group’s total errors, and 12.53% of the TH group’s total errors. No significant differences between groups were found for the proportions of either ‘1’ scores ($p = 0.109$) or ‘0’ scores ($p = 0.719$).

3.4.2.2. Regression analyses. Spelling scores for both irregular words and nonwords represented the outcome variables for all regression analyses performed. Predictor variables included orthographic processing (OP), phonological awareness (PA), phonological memory (PM) and letter-sound knowledge (LSK). Since the latter three predictors from the above list measured phonological processing, it was not considered appropriate to include them in the one regression model, given the potential for collinearity effects. Three regression models were therefore computed, containing OP and each of the phonological measures separately. As with the group comparison, nonword spelling data for one TH participant were excluded from regression analyses, due to concerns about outlier effects.

The results of the regression analyses for irregular words and nonwords are summarised in Tables 3 and 4, respectively. For both groups and in all multivariate regression models, irregular word spelling (Table 3.3) was predicted by orthographic processing only. The concurrent (i.e., adjusted) contributions of phonological processing did not reach significance. In addition, the amount of irregular word spelling variance explained by each of the models was similar for the two groups (CI: $R^2_{adj} = 0.605 – 0.687$; TH: $R^2_{adj} = 0.552 – 0.568$).

A different pattern of relationships was revealed for nonword spelling (Table 3.4). For the CI group, nonword spelling variance was explained only by PA, both when it was in isolation ($R^2_{adj} = 0.321$) and when it was combined with OP ($R^2_{adj} = 0.308$). Hence, for the CI group, a substantial amount of nonword spelling variance may have been contributed by factors outside of those selected to be predictor variables. This finding contrasted with the TH group, for whom the regression model with the most explained variance included both LSK and OP ($R^2_{adj} = 0.625$). The contribution of LSK was statistically significant here, though that of OP was not.

3.4.2.2.1. Power analyses. A post hoc analysis was carried out with the CI group, in order to determine the statistical power available for multiple regression analyses with 14 participants and two predictor variables. Results indicated that the power value required to find a squared multiple correlation coefficient of medium size (i.e., 0.5) was 0.65. This value was considered acceptable, although a limitation of the study was that there may have been insufficient power to find smaller multiple correlation coefficient values. Where the regression results indicate a non-significant relationship between predictor and outcome variables, the possibility of a Type II error should therefore be considered. That said, the regression analyses for irregular and nonsense word spelling did return some significant results, suggesting that these observed relationships were resistant to Type II errors.

Table 3.3

Multiple regression results for irregular word spelling.

	CI (n=14)				TH (n=31)			
	R^2_{adj}	β	*t*	*p*	R^2_{adj}	β	*t*	*p*
Unadjusted								
OP	.637	.815	4.878	*<.001	.559	.758	6.250	*<.001
PA	-.054	.165	.581	.572	.126	.394	2.307	*.028
PM	-.072	.101	.350	.732	.156	.429	2.557	*.016
LSK	-.033	.215	.763	.460	.370	.626	4.247	*<.001
Adjusted	*.614				*.565			
OP		.847	4.669	*.001		.705	5.507	*<.001
PA		-.100	-.550	.593		.152	1.189	.244
Adjusted	*.687				*.552			
OP		.938	5.487	*<.001		.711	5.163	*<.001
PM		-.292	-1.709	.116		.101	.734	.469
Adjusted	*.605				*.568			
OP		.807	4.520	*.001		.598	3.718	*.001
LSK		.038	.214	.834		.236	1.466	.154

Note. CI = cochlear implant; TH = typically hearing; OP = orthographic processing; PA = phonological awareness; PM = phonological memory; LSK = letter-sound knowledge. Statistically significant values (p<.05) indicated with asterisk (*).

Table 3.4

Multiple regression results for nonword spelling.

	CI (n=14)				TH (n=29)			
	R^2_{adj}	β	*t*	*p*	R^2_{adj}	β	*t*	*p*
Unadjusted								
OP	.077	.385	1.444	.174	.409	.656	4.517	*<.001
PA	.321	.611	2.674	*.020	.203	.481	2.851	*.008
PM	.078	.387	1.452	.172	.094	.355	1.975	.059
LSK	.094	.405	1.534	.151	.596	.781	6.501	*<.001
Adjusted	.308				*.450			
OP		.214	.883	.396		.552	3.624	.001
PA		.544	2.240	*.047		.264	1.732	.095
Adjusted	.066				*.388			
OP		.270	.915	.380		.638	3.736	*.001
PM		.273	.926	.374		.036	.210	.835
Adjusted	.121				*.625			
OP		.311	1.166	.268		.266	1.771	.088
LSK		.337	1.263	.233		.611	4.070	*<.001

Note. CI = cochlear implant; TH = typically hearing; OP = orthographic processing; PA = phonological awareness; PM = phonological memory; LSK = letter-sound knowledge. Statistically significant values (p<.05) indicated with asterisk (*).[1]n=29 for TH group due to missing values for one participant and removal of one outlier.

3.5. Discussion

The present study examined spelling in young children with hearing loss who used spoken communication and CIs. The results of this cohort were compared with a TH group of children, in terms of: (1) the absolute accuracy of irregular and nonsense word spelling results, and; (2) the contribution of a range of underlying processing skills to spelling outcomes. Contrary to expectations, spelling accuracy was similar across groups. Children with CIs, however, showed underlying phonological processing difficulties, and evidence from error and regression analyses suggested that they relied less on phonics-based skills to produce written words.

3.5.1. Spelling accuracy in children with cochlear implants. Children with CIs demonstrated similar irregular word and nonword spelling accuracy, when compared with their hearing peers. This outcome did not align with results from prior studies that have reported significant spelling difficulties in children with hearing loss, both with CIs (Apel & Masterson, 2015; Geers & Hayes, 2010; Roy et al., 2015) and without (Arfé et al., 2015; Harris & Moreno, 2004; Nelson & Crumpton, 2015; Roy et al., 2015). That said, the findings are not altogether exceptional. Fitzpatrick and colleagues (2012) found that 80% of children with CIs performed within normal limits on a standardised measure of real word spelling. Similarly, Hayes, Kessler and Treiman (2011) also reported no statistically significant group differences in spelling accuracy between CI and chronological age-matched hearing children, although it is unclear if Hayes and colleagues' results would have differed if the children's reading comprehension ability had not been statistically controlled.

It is possible that our CI cohort's comparatively better spelling accuracy outcomes are attributable to use of a spoken communication mode. Of the studies listed above, those that included strictly spoken language communicators (e.g., Fitzpatrick et al., 2012; Hayes et al., 2011) tended to report better outcomes than those that included children from a range of communicative backgrounds (e.g., Apel & Masterson, 2015; Roy et al., 2015). Children in the present study used only spoken language and were exposed from a young age to AVT, which emphasises the use of speech to communicate. Hence, the relatively successful spelling outcomes obtained by the group may be associated with their consistent use of spoken language, although further research that directly compares children from different communication backgrounds would be needed to confirm this assertion.

To date, the present study is the first to have used both irregular word and nonword stimuli to measure spelling accuracy in children with CIs. This approach allowed for different interpretations to be made, based on the CI cohort's performance on each of the two word types. When spelling a nonsense word, an examinee must analyse the item's phonemic structure, in order to accurately map the appropriate sequence of letters to speech sound constituents (Curtin, Manis & Seidenberg, 2001). Hence, nonword spelling depends more directly on phonological processing than does irregular word spelling, because by definition, irregular words contain uncommon letter-sound correspondences which must – at least partially – be coded using knowledge of existing orthographic information (Houghton & Zorzi, 2003). Children with and without CIs in the present study demonstrated similar accuracy on the irregular word spelling task, indicating that the successful retrieval of items' orthographic representations was not significantly influenced by hearing status. The CI and TH groups also demonstrated statistically similar results when spelling nonwords, although given the trend towards a significant group difference ($p = 0.064$) and the absence of existing evidence regarding the nonword spelling abilities of children with CIs, further research is warranted to explore this finding.

It is possible that, given the exact demands of the nonword spelling task, the CI group may have had difficulty perceiving nonword stimuli as clearly as the TH group. All items were presumably unfamiliar to the participants, which means that good speech discrimination skills would have been critical to spelling performance. The obvious factor to consider is that children with CIs perceive speech stimuli via an external hearing device, rather than a fully functioning cochlea. Future researchers using a nonword spelling task with this population may consider taking additional steps to control for speech perception difficulties, such as having participants repeat the target word before producing it in writing.

Yet, despite the potential for failure at an auditory perceptual level, the testing conditions for nonword spelling in the present study were such that this explanation is unlikely. Each item in the task was clearly articulated to participants at least twice via face-to-face speech, in an environment with no background noise. Additionally, all children were educated in a mainstream setting and they used only spoken language to communicate. The findings were therefore considered to reflect the functioning of underlying linguistic processors and not the sensory perception of input at the time of testing. This position is supported by research findings that nonword *reading* is also found to be a difficult task for children with CIs, presumably due to the phonological processing demands associated with decoding the speech sounds represented by an unfamiliar letter sequence (Bouton, Colé, Serniclaes, Duncan & Giraud, 2015; Geers & Hayes, 2010; Weisi et al., 2013).

3.5.2. Skills underlying spelling accuracy in children with cochlear implants. In addition to evaluating the absolute spelling accuracy of children with CIs, the present study analysed the skills on which spelling outcomes were based. Firstly, the spelling errors made by all individuals were examined, with reference to the types of errors made, and the graded accuracy of these errors at phoneme and word levels. On average, 36.11% incorrect words produced by the CI group were phonologically plausible (i.e., homophones or pseudohomophones). This proportion was significantly lower than the TH group, for whom 48.59% incorrect words were phonologically plausible. The group difference was predicted based on similar findings reported in the literature (Harris & Terlektsi, 2011; Hayes et al., 2011; Roy et al., 2015). Interestingly, the exact proportional values for phonologically plausible errors reported in previous studies are 44% for 9-year-olds (Hayes et al., 2011), 52% for 11-year-olds (Roy et al., 2015), and 62% for adolescents (Geers & Hayes, 2010). Thus, the percentage of phonologically plausible errors produced by children aged 6 to 9 years with CIs in the present study fits well into a developmentally appropriate pattern of increasing phonological sensitivity with age. An alternative explanation is that the lower proportion of phonologically plausible errors reported here is attributable to other variables, such as communication mode or age at cochlear implantation. Yet, our cohort was similar to those examined by Hayes and colleagues (2011) with respect to oral language and CI use, and they received their implants at a younger age than Hayes et al.'s group. It is thus not immediately clear what factor aside from age at testing would account for reduced phonological plausibility found in the present study, relative to what has been previously reported.

The Spelling Sensitivity Score (SSS) system used to explore written word productions in the present study allowed for an examination of errors at both 'element' (i.e., phoneme and morpheme) and word levels. While children with CIs obtained a lower average SSS-Element (SSS-E) score than TH children, the same divergence between groups failed to reach statistical significance for SSS-Word (SSS-W) scores. This is likely attributable to the different degrees of sensitivity that pertain to the two scores. The SSS-W yields a single score for each item, whereas the SSS-E takes account of all phonemes and morphemes in a given word, so there are more opportunities for differences to emerge between groups. Based on the entire error profile found for each group, inaccurate words produced by TH children included more plausibly – though incorrectly – represented elements, which therefore retained the item's phonology (e.g., 'sckool' for 'school'). In comparison, elements in words produced by children with CIs were misspelled such that the entire item tended to be less phonologically plausible (e.g., 'goot' for 'good').

Further analyses were conducted to explore the potential underlying difficulties that characterised phonologically implausible misspellings in children with CIs. An important aspect of the SSS system is that element omissions are scored differently to illegal letter-sound substitutions, although both error types are phonologically implausible in a dichotomous system of categorisation. According to Masterson and Apel (2010), element omission errors represent phonemic awareness limitations, whereas illegal letter substitutions represent reduced knowledge of letter-sound (or letter-affix) correspondences. The former error type therefore relates to phonological awareness, while the latter relates more specifically to phonics skills.

The proportion of errors in which an element was omitted in misspelled words was roughly equivalent across groups in the present study (CI = 13.19%, TH = 12.53%). Similarly, the difference between CI and TH children was not statistically significant in terms of illegal letter-sound substitution errors (CI = 50.69%, TH = 38.87%). Hence, the distribution of scores across the two implausible error types was such that, although phonological plausibility overall was reduced for children with CIs, the exact source of this divergence could not be confirmed. Further research is warranted to investigate this question in depth, since many linguistic processes apart from phonemic awareness contribute to written word spelling development (Apel & Masterson, 2015). Certainly, the finding that children with CIs produced a similar proportion of phonemic omission errors to TH children indicates that they were largely aware of target words' phonemic structure, although they may have had difficulty applying that knowledge to represent phonemes or morphemes with letters.

Importantly, the group differences described above are based only on an examination of inaccurate word productions. To evaluate the factors contributing to irregular word spelling accuracy more broadly, regression analyses were computed, which included phonological and orthographic processing skills as predictor variables. The profiles were very similar between CI and TH groups: orthographic processing alone contributed significantly to irregular word spelling. This pattern of results makes theoretical sense, since irregular words contain uncommon letter-sound correspondences that rely on whole or partial retrieval of an item’s existing orthographic representation (Houghton & Zorzi, 2003).

At first glance, the regression findings for irregular word spelling appear at odds with the error analysis. As described, the regression outcomes suggest that all children, both with and without CIs, used primarily orthographic processing to spell irregular words successfully. Yet, inaccurate words produced in the same test by the TH group were more phonologically plausible than those for the CI group, indicating that children with typical hearing relied more so on a phonics-based method of coding items when the target word was difficult enough to have elicited an error. Together, the results from regression and error analyses therefore point to the two groups using different real word spelling strategies, whereby children with CIs tend to apply less phonics knowledge to ‘sound out’ an item for which they do not have an existing orthographic representation. Indeed, a similarly reduced application of phonics knowledge was also inferred from the CI group’s nonword spelling regression findings.

Nonword spelling accuracy for the TH group was explained best by a regression model combining letter-sound knowledge and orthographic processing (although the contribution of orthographic processing in the model was not statistically significant). The association with letter-sound knowledge is understandable, since in order to correctly code the pronunciation of a target nonword, the speller needs to apply their knowledge of how phonemes are represented by certain letters and letter sequences. In contrast, letter-sound knowledge did not contribute significant variance to nonword spelling for children with CIs. This finding suggests that the group did not apply phonics knowledge to the same degree as TH children during the task. In addition, their comparatively increased proportion of implausible errors with irregular words supports the position that children with CIs may apply phonics knowledge less effectively than TH children when spelling more generally.

The only variable to contribute significant variance to nonword spelling accuracy in the CI group was phonological awareness, which indicates that children with CIs were able to access items’ phonemic structure using the same explicit knowledge required for completing phonological awareness tasks, such as omitting, blending or identifying parts of spoken words. Again, this finding aligns with results from the irregular word spelling error analysis, in which phonemes were not usually omitted in inaccurate spellings produced by either group.

In contrast with our results however, Nelson and Crumpton (2015) did not find a significant relationship between phonemic awareness and nonword spelling accuracy in children with hearing loss aged 6 to 18 years old. Given the early age at which phonological (and hence, phonemic) awareness skills are generally acquired, the variability in performance is expected to be greater in our comparatively younger cohort (Luft, 2018). It is then perhaps unsurprising that there was a significant relationship found between phonological awareness and nonword spelling in the present study, but not in the study by Nelson and Crumpton (2015). Moreover, our cohort with hearing loss was homogeneous in terms of communication mode and use of CI technology, and the results reported here may therefore better represent this subset of the population, compared with those pertaining to the far more heterogeneous group examined by Nelson and Crumpton (2015).

It may be noted that children with CIs in the present study still demonstrated phonological awareness difficulties, despite the finding that these skills related significantly to nonword spelling accuracy. Hence, other underlying skills must have contributed to the group's relative success with nonword spelling, and indeed the total amount of nonword spelling variance contributed by phonological awareness in isolation was 32.1%, which leaves a large amount of unexplained variance that could not be accounted for by the selected predictor variables. Further research is required to expand on the present study's findings and examine other potential variables that might influence nonword (i.e., unfamiliar word) spelling outcomes.

The cohort with hearing loss in the present study was relatively homogeneous, since they all had bilateral hearing loss, used CIs and spoken communication, were exposed early in life to AVT, and were in the first three years of formal literacy instruction. Still, the children would also have had different written and spoken language experiences in the home and at school, and these were not examined here. The children may also have varied in the degree to which visual processing of phonological information contributed to outcomes, such that some children's spelling performances were possibly facilitated by implicit articulatory cues. It would be interesting for future studies to manipulate the presentation characteristics of nonword stimuli, and to otherwise explore the individual and demographic factors that influence spelling development in children with hearing loss.

3.6. Conclusion

The present study is the first to compare the spelling skills and sub-skills of young children with CIs, who communicate via spoken language, with those of typically hearing children of the same chronological and mental age. The children with CIs demonstrated similar irregular word spelling accuracy, and while they appeared to find nonword spelling more challenging, results were again statistically similar between groups. Taken together, results of error and regression analyses point to key processing differences between the two groups. More specifically, children with CIs appeared less able to apply phonics strategies when spelling, in comparison to their hearing peers. It would be interesting for future research to extend on the present study's cross-sectional analyses, by exploring the longitudinal spelling outcomes of spoken language users with CIs. In particular, different types of literacy-based instruction may be compared, in order to determine how best to support this cohort's development in the long term.

Chapter 4.

Semantic Processing in Children with Cochlear Implants: Evidence from Event-related Potentials

Chapters 2 and 3 presented a comprehensive examination of literacy outcomes in young children with cochlear implants, as measured by their behavioural assessment responses. The findings were intended to represent how this subset of the population might perform on real-life reading and spelling tasks within an educational or clinical setting. The behavioural responses themselves, however, do not provide specific information about the functioning of underlying neural processes that contribute to literacy development. A reader's recognition of a given word – either spoken or presented in print – is influenced by their processing of that word's semantic properties. Similarly, semantic processing abilities contribute to comprehension of higher level sentence- and passage-level information, as a word's meaning is integrated with the surrounding context. In Chapter 4, electroencephalography (EEG) will be used to capture participants' sensitivity to lexical-semantic incongruence, on a fine-grained temporal scale. The results are intended to provide insight into how children with cochlear implants process semantic information on a neural level, the implications of which are critical in gaining a more complete understanding of how, more broadly, literacy skills develop in this population.

Chapter 4 presents a corrected version of an article submitted for journal publication[1], which is currently under review. Minor changes have been made to formatting and methodological description, in order to maintain thesis continuity. The authors' contributions to the original submitted article are detailed on the following page.

[1] Bell, N., Angwin, A.J., Arnott, W.L., & Wilson, W.J. (2018*). Semantic processing in children with cochlear implants: evidence from event-related potentials.* Manuscript under review.

Contributor	**Statement of contribution**
Nicola Bell (Candidate)	Conception and design: 65% EEG task preparation: 90% Data collection: 100% Data analysis: 70% Interpretation: 80% Manuscript writing: 100% Manuscript revisions: 20%
Dr Anthony Angwin	Conception and design: 15% EEG task preparation: 5% Data analysis: 15% Interpretation: 10% Manuscript revisions: 30%
Dr Wendy Arnott	Conception and design: 10% Interpretation: 5% Manuscript revisions: 20%
A/Prof Wayne Wilson	Conception and design: 5% EEG task preparation: 5% Data analysis: 15% Interpretation: 5% Manuscript revisions: 30%

4.1. Abstract

The present study sought to determine whether processing of lexical-semantic incongruence, as indexed by the N400 effect, differed between children with cochlear implants (CIs; n = 12) and a typically hearing (TH) control group (n = 30). Event-related potentials (ERPs) were recorded while participants, aged 6-9 years, responded to a spoken word-picture matching task. A significant N400 effect was elicited in both groups. Despite the similarity in ERP responses, children with CIs scored significantly lower on behavioural measures of language and reading. No significant correlations between ERP and behavioural measures were found, although there was a trending relationship between sentence-level spoken language comprehension and the TH group's N400 effect mean amplitude. The results suggest that, at a neural level, children with CIs can process lexical-semantic incongruence, and that other underlying processes not measured by the N400 effect contribute to this population's language and literacy difficulties.

4.2. Introduction

Children with varying degrees of hearing loss are generally found to have spoken and written language difficulties, relative to their typically hearing peers (e.g., Nelson & Crumpton, 2015). Advances in hearing technology mean that even those children with a profound hearing loss can access spoken communication via cochlear implants (CIs). However, while the early speech and language learning trajectories of young children are reportedly enhanced following cochlear implantation (Tait, Nikolopoulos & Lutman, 2007), a discrepancy still remains between the spoken language comprehension skills of school-age children with CIs and those of typically hearing children (Ceh, Bervinchak & Francis, 2013; Ching, Day & Cupples, 2014; Fitzpatrick et al., 2012; Spencer, Barker & Tomblin, 2003). Language difficulties observed in children with CIs are also posited to result in disordered literacy development (Spencer et al., 2003). Indeed, for typically hearing students, spoken language skills are found to influence word-level reading development (Walley, Metsala & Garlock, 2003), as well as the overall comprehension of written text (Catts, Adlof & Weismer, 2006; Hoover & Gough, 1990).

Existing research pertaining to the spoken and written language skills of children with CIs has, for the most part, been conducted using behavioural testing methods, with few studies examining the specific neural processes that underlie language and literacy performance. Information obtained with behavioural assessments is valuable, as the results may be generalised and applied to real-life observations. However, responses to such assessments also represent the end-point of a complex network of underlying cognitive processes (Henderson, Baseler, Clarke, Watson & Snowling, 2011). Hence, behavioural test results may be complemented and expanded upon by evidence from direct measures of neurological function, such as that obtained via electroencephalography (EEG). The present study used both EEG and behavioural testing to examine the lexical-semantic processes that underlie spoken language and reading comprehension in beginning readers with CIs.

In past research, EEG technology has been used in combination with experimental paradigms to elicit event-related potentials (ERPs). Event-related potentials are thought to indicate changes in time-locked post-synaptic activity following exposure to a stimulus (Kutas, Van Petten & Kluender, 2003). Different ERP 'components' may be extracted from EEG activity, and are generally characterised by the polarity and number of peaks contained in each ERP, the latency with which these peaks occur, and the distribution of ERP activity across the scalp (Kotz & Friederici, 2003). The precise temporal resolution of ERP measurement makes it ideal for studying language processing, which itself is founded on numerous underlying mechanisms occurring over a short time period. In the specific context of investigating semantic processing, the most commonly considered ERP component is the N400 (Kutas et al., 2003).

The N400 component is displayed as a negative waveform peak, occurring at approximately 400 milliseconds after stimulus onset (Kotz & Friederici, 2003). The amplitude of the N400 is especially sensitive to the predictability of linguistic context. More specifically, the greater the extent to which word retrieval is facilitated by lexical or semantic factors, the less negative the waveform amplitude is in response (Kutas & Federmeier, 2011). The difference in amplitude elicited by semantically predictable relative to unpredictable stimuli is termed the 'N400 *effect*'. This effect is frequently examined as an index of semantic retrieval and lexical-semantic integration (Kotz & Friederici, 2003). Sensitivity to lexical-semantic incongruence, as represented by a significant N400 effect, has been demonstrated by typically developing children as young as 5 years of age (Byrne et al., 1999).

Generally speaking, the ability to process semantic information is fundamental among the factors contributing to overall language comprehension (Harm & Seidenberg, 2004). In terms of behavioural assessment measures, those that tap into vocabulary awareness are presumed to have high semantic processing demands, since responses to such tasks are inherently dependent on knowledge of word meanings isolated from context (Stites & Laszlo, 2017). Of course, context is key to language comprehension at a discourse level, which means that semantic processing is also required for multi-item comprehension tasks wherein a word's semantic representation is integrated with information from the rest of the sentence (Van Berkum, Brown, Zwitserlood, Kooijman & Hagoort, 2005).

The link between behavioural and ERP measures of semantic processing has previously been examined in research involving typically hearing populations. Henderson and colleagues (2011) elicited ERP responses in 8- to 10-year-old children, using a passive picture-word task. An N400 effect was obtained, as defined by a significantly more negative peak amplitude for the incongruent picture-word pairs compared to congruent pairs. The size of the effect was correlated significantly with behavioural performance on a passage-level spoken language comprehension task. Interestingly, no correlation was found between the N400 effect and word-level receptive vocabulary. Based on their results, the authors suggested that the N400 effect appeared to reflect – at a neural level – the ease with which individuals integrated semantic information with contextual information to achieve comprehension (Henderson et al., 2011).

Deviant N400 effects in certain clinical populations have also been reported in the ERP literature. Cummings and Ceponiene (2010) used a picture-word matching paradigm to explore semantic processing in 7- to 15-year-old children with specific language impairment (SLI). Although the mean amplitude of the N400 effect was similar for this group and a group of typically developing age-matched controls, the peak latency was significantly delayed in children with SLI. The authors suggested that this delay was representative of semantic integration deficits, as observed in behavioural assessments of the group’s spoken language skills. Similarly, reduced N400 effect amplitudes have been found for dyslexic children (Schulz et al., 2009) and delayed N400 effect latencies have been found for dyslexic adults (Helenius, Salmelin, Service & Connolly, 1999).

Based on the above results involving other clinical populations, it may be predicted that individuals with CIs will show different N400 effects when compared with typically hearing controls. There are only a few studies, however, that have addressed this question. With respect to adults with CIs, Hahne, Wolf, Müller, Mürbe and Friederici (2012) examined the N400 effect using a cloze sentence task to elicit ERP responses. The mean amplitude of the N400 effect was similar for the CI group and typically hearing adults. However, for the CI group only, the effect was extended over a longer post-target time period, which the authors attributed to delayed semantic integration processes in adults with CIs (Hahne et al., 2012).

Compared with the literature on adult CI users, relatively more research has been conducted to investigate the N400 effect in children with CIs (Key, Porter and Bradham, 2010; Kallioinen et al., 2016; Vavatzanidis, Murbe, Friederici & Hahne, 2018), although only one study to the author’s knowledge has addressed this evidence gap on a group scale and with reference to typically hearing controls. Kallioinen and colleagues (2016) elicited ERP responses in a 5- to 7-year-old CI cohort, who used a combination of sign and spoken language. The study’s word-picture matching task included three different conditions: stimulus pairs were either congruent (e.g., wolf-wolf), between-category incongruent (e.g., wolf-car), or within-category incongruent (e.g., wolf-bear). In response to the ‘within-category incongruent’ condition, children with CIs produced similar mean N400 effect amplitudes to typically hearing children and children with hearing aids. Unexpectedly however, the mean amplitude of the N400 effect associated with ‘between-category incongruence' was significantly larger for the CI group relative to the other groups. This result was not interpreted as indicating enhanced semantic sensitivity, since the group with CIs performed below the standard of their peers on a behavioural lexical access task administered as part of the same study. Instead, the finding was attributed to different processing strategies and levels of motivation between groups.

A related and potentially confounding factor in the study by Kallioinen and colleagues (2016) was the weighting of word-picture stimuli. Given that there were two incongruent conditions, only a third of the total number of word-picture pairs were matching. The authors suggested that the control group children may have realised that the target stimulus was not predicted by its prime in the majority of trials, and so used a more passive, bottom-up mode of processing, which in turn reduced the overall ERP effects of semantic relatedness. In contrast, since the task appeared more challenging for children with CIs, this group may have continued to implement a predictive processing strategy, which thus had a different influence on the size of N400 effect. Given the difficulty in interpreting these results, there is a need for additional neurophysiological research that uses an ERP approach to examine how children with CIs access and integrate lexical-semantic information. Such evidence about language processing, captured on-line and in real time, would complement the already abundant behavioural research that demonstrates this population's spoken and written language difficulties (e.g., Geers, 2003; Nittrouer, Caldwell, Lowenstein, Tarr & Holloman, 2012; Nittrouer, Caldwell-Tarr, Sansom, Twersky & Lowenstein, 2014; Weisi et al., 2013).

In addition to examining the N400 effect in children with CIs, Kallioinen and colleagues (2016) included behavioural assessments of phonological processing, cloze sentence completion and reading. No correlations were found between these measures and the between-category N400 effect amplitude, but a significant correlation was observed between the within-category N400 effect amplitude and cloze sentence completion performance – a finding which may highlight the N400's sensitivity to word expectancy violations (Kutas & Federmeier, 2011). The amplitude of the within-category N400 effect was also correlated with phonological processing, although this finding cannot easily be explained by shared lexical-semantic demands across ERP and behavioural tasks. Most importantly, Kallioinen et al. (2016) did not include behavioural assessments of spoken language comprehension. Accordingly, it is not possible to know the degree to which the language difficulties exhibited by children with CIs relate to their underlying lexical-semantic functioning, as indexed by the N400 effect. Further insight may be obtained by examining the correlations between ERP and behavioural language measures in this cohort, as has been done in typically hearing children (e.g., Henderson et al., 2011).

4.2.1. Current study. To date, substantial behavioural research has highlighted the spoken and written language difficulties experienced by children with CIs. Yet, the same aims have rarely been addressed using neurophysiological methods, and with attention to the N400 effect. Where CI users have been the focus of investigation, the N400 effect is reportedly significant (Hahne et al., 2012; Kallioinen et al., 2016; Key et al., 2010; Vavatzanidis et al., 2018), albeit with a different waveform shape to that elicited in typically hearing controls (Hahne et al., 2012; Kallioinen et al., 2016). Of the previous studies involving children with CIs, none has examined the N400 exclusively in users of spoken language. This factor makes interpretation of results difficult, since communication mode is associated with different behavioural literacy results in children with aided hearing loss (Cupples et al., 2017; Johnson & Goswami, 2010; O'Donoghue, Nikopoulos & Archbold, 2000). Moreover, length of experience with using sign language has been found to influence the N400 effect in hearing adults (Zachau et al., 2014). The only prior study to have conducted group-level comparisons of the N400 effect in children with CIs (i.e., Kallioinen et al., 2016) included 5-year-old participants, who may therefore be too young to be considered 'readers'. Based on the state of existing research literature, the status of the N400 effect in children with CIs who are solely spoken communicators, and whose reading skills are developing with exposure to formal literacy instruction, remains unclear.

In an effort to address these gaps in evidence, the first research question in the present study asked: Is semantic processing, as indexed by the mean amplitude and peak latency of the N400 effect, altered in beginning readers with CIs who communicate via spoken language? The word-picture matching task used to elicit the N400 effect included an equal number of matching and non-matching stimuli. Given the lack of existing N400-related literature involving children with any degree of hearing loss, all current analyses were considered exploratory; however, based on evidence of relatively poor performance by this population on behavioural measures of vocabulary and listening comprehension (e.g., Ching et al., 2014; Johnson & Goswami, 2010), it was tentatively predicted that the N400 effect elicited in the CI group would be reduced in amplitude and/or delayed, compared with typically hearing children.

A secondary aim of this study was to determine whether the N400 represented a neural marker of language and reading for both typically hearing children and children with CIs. Specifically, the present study asked: Are behavioural spoken language and reading outcomes related to the amplitude or latency of the N400 effect, and does the strength of this relationship differ in children with CIs?

4.3. Method

4.3.1. Participants. Twelve children with CIs (3 left-handed, 6 females) and 30 typically hearing (TH) children (3 left-handed, 15 females) participated in the present study. Most children with CIs and many of the typically hearing participants were recruited from Hear and Say, an organisation that provides audiological and spoken language intervention to children with hearing loss, as well as conducting regular school-based hearing screening. Parents and caregivers who had given permission to be contacted for research purposes were invited to participate in the study. The remaining participants were recruited from the wider community via newsletter advertisements and word of mouth.

All participants adhered to the following inclusion criteria: (1) use of spoken English as native and primary form of communication; (2) nonverbal reasoning at or above normal limits (as measured using the *Raven's Coloured Progressive Matrices*; Cotton et al., 2005; Raven, Raven & Court, 2004); (3) no diagnosed developmental disorders or intellectual disabilities, and; (4) in Grade 1, 2 or 3 of a mainstream school. The CI and TH groups did not differ on gender, $\chi^2(1) = 0.038$, $p = 0.845$; handedness, $\chi^2(1) = 1.575$, $p = 0.209$; age, $t(40) = -1.525$, $p = 0.135$; grade, $\chi^2(2) = 1.610$, $p = 0.447$, or nonverbal reasoning, $t(40) = 0.780$, $p = 0.440$. With regard to nonverbal reasoning, 'normal limits' was operationally defined as performance within one standard deviation of the mean (i.e., a z-score on the *Raven's Coloured Progressive Matrices* of between -1 and +1). Age and nonverbal reasoning scores were checked to confirm that data were normally distributed within each group and variances were equal between groups. There were no outliers for age or nonverbal reasoning in either group.

Additional inclusion criteria for the children with CIs were also implemented: participants needed to have received their CIs before the age of 5 years, and have received AVT before entering formal schooling. In all cases, AVT was administered by AVT-certified speech pathologists or AVT-certified Teachers of the Deaf. The mean age of the CI group at the time of testing was 8.10 years (SD = 0.85y). These participants were in Grade 1 (n = 1), Grade 2 (n = 5) or Grade 3 (n = 6). The CI group's average nonverbal reasoning z-score was 0.64 (SD = 1.28). All children received CI surgery before the age of 5 years (mean = 1.93y; SD = 1.48y), and the mean duration of CI use at the point of EEG testing was 6.12 years (SD = 1.54y). The mean age at enrolment in early intervention was 10 months (median = 0.29y; range = 0.08-3.25y). Information pertaining to each participant is shown in Table 4.1 (see Chapter 2 of this thesis for additional details relating to the CI cohort).

Participants in the TH control group passed hearing screening assessments (thresholds of 25dB HL or better at octave intervals of 500 to 4000 Hz) conducted using a commercially available screening audiometer. The participants completed these screenings after entering formal schooling and no more than two years prior to their participation in the present study. The mean age of the TH group was 7.68 years (SD = 0.77y). Five participants were in Grade 1, 16 were in Grade 2, and 9 were in Grade 3 at the time of testing. The group's average nonverbal reasoning z-score on the *Raven's Coloured Progressive Matrices* was 0.93 (SD = 0.99).

4.3.2. Ethics statement. Ethical approval for the present study was obtained from the Behavioural and Social Sciences Ethical Review Committee at the University of Queensland. Gatekeeper ethical approval was also obtained from the Hear and Say Research and Ethical Advisory Committee. All parents gave written informed consent for their children to participate in the study.

Table 4.1

Audiometric information for participants with CIs (n=12).

#	Configuration	Aetiology	Stable (Y/N)	Age (m) at 1st implant	Age (m) at EI enrolment	Unaided PTA (dB)		Aided PTA (dB)	
						Left	Right	Left	Right
1	CI+CI	Con. (idiopathic)	Y	46	2	75.00	76.25	25.00	25.00
2	CI+CI	Con. (idiopathic)	N	37	29	75.00	63.75	22.50	22.50
3	CI+CI	Con. (idiopathic)	N	31	3	88.75	82.50	22.50	18.75
4	CI+CI	Con. (idiopathic; LVAS)	Y	9	4	76.25	97.50	21.25	22.50
5	CI+CI	Con. (genetic-NS)	Y	8	2	98.75	98.75	25.00	22.50
6	CI+CI	Con. (genetic-Connexin 26)	Y	7	1	≥100	≥100	22.50	18.75
7	CI+CI	Con. (genetic-Connexin 26)	Y	8	1	95.00	≥100	22.50	21.25
8	CI+CI	Acq. (1;3y; idiopathic)	Y	19	31	≥100	≥100	27.50	22.50
9	CI+CI	Con. (CMV)	Y	8	4	≥100	98.75	25.00	21.25
10	CI+CI	Con. (CMV)	Y	8	7	97.50	≥100	30.00	28.75
11	CI+HA	Con. (genetic-Pendred Syndrome)	N	47	1	96.25	75.00	28.75	*
12	HA+CI	Acq. (1;3y; idiopathic; LVAS)	N	50	39	61.25	77.50	30.00	30.00

Note. CI = cochlear implant; HA = hearing aid; EI = early intervention; PTA = pure tone average; Con. = congenital; Acq. = acquired; LVAS = Large Vestibular Aqueduct Syndrome; NS = not specified; CMV = Cytomegalovirus. *Data for left ear missing but reportedly similar to right ear.

4.3.3. Behavioural testing. Behavioural measures were administered by a qualified speech pathologist prior to EEG testing and usually on a separate day, as part of a larger behavioural assessment battery. Where any of the above behavioural tasks were completed on the same day as EEG testing, their duration did not exceed 10 minutes, and participants were provided with a break before beginning EEG testing.

Word-level reading accuracy was assessed using the *Castles and Coltheart 2* (*CC2;* Castles et al., 2009*),* a standardised test normed for Australian children in Grades 1 through 6 (i.e., age 6 through 11;6 years). Examinees are asked to read aloud three different word types: (1) nonwords, which have regular spellings but are not real words (e.g., ‘gop’); (2) irregular words, which have irregular English phoneme-grapheme correspondences (e.g., ‘good’); (3) regular words, which have regular English phoneme-grapheme correspondences (e.g., ‘bed’). In total, there are 120 items of increasing difficulty (40 of each word type), and each item is presented on an individual card. The examinee is scored 1 for an item if their response accurately reflects the standard adult pronunciation. A score out of 40 for each word type was obtained and this was converted to a standardised z-score, using the test norms. Word reading skill in the present study was represented by a composite score, calculated as the average of each participant’s regular word, irregular word and nonword reading accuracy standard scores on the *CC2*.

In addition to word-level reading tasks, participants’ passage reading comprehension was assessed using the *Primary Passage* subtest of the *York Assessment of Reading for Comprehension – Australian edition* (*YARC*; Snowling et al., 2012). Performances were scored using the test’s standardised Australian norms for children aged 5 to 12 years. In this assessment measure, children read aloud two passages of text selected according to their age and accuracy level. They then answer approximately eight questions about the passage, which require the use of both literal and inferential comprehension skills. Children receive a score of either ‘0’ or ‘1’ on the basis of their accuracy when answering questions, and their total raw score – from which the standard score is derived – is calculated as the cumulative total.

Receptive vocabulary was assessed using the *Peabody Picture Vocabulary Test – 4th edition* (*PPVT-IV*; Dunn & Dunn, 2007). Standardised norms are available for individuals over the age of 2;6 years. In the *PPVT-IV*, examinees are presented with four pictures and are asked to point to the one that corresponds with a word spoken by the test administrator. There are 228 items in total, composed of 19 equally distributed and increasingly difficult item-sets.

The test is widely used as a measure of receptive vocabulary, or the ability to understand the meaning of single words in isolation. Raw scores were converted to standard scores.

Receptive language skills were also assessed at the sentence and passage levels, using two subtests from the *Clinical Evaluation of Language Fundamentals – 4th edition* (*CELF-4*; Semel, Wiig & Secord, 2003). 'Sentence-level comprehension' was defined in the present study as performance on the 'Concepts and Following Directions' subtest of the *CELF-4*. In this task, examinees are required to carry out directions that contain increasingly advanced concepts within increasingly long and complex sentences. 'Passage-level comprehension' was defined as performance on the 'Understanding Spoken Paragraphs' *CELF-4* subtest. Here, participants were assessed on their ability to answer comprehension questions about a short text read aloud to them. Scaled scores (with normative mean scores of 10 and standard deviations of 3) from each of these subtests were included in statistical analyses.

4.3.4. ERP testing.

4.3.4.1. *Picture stimuli.* Stimuli for the ERP testing consisted of 60 coloured photographs, paired with 60 congruent spoken word labels and 60 incongruent spoken word labels. In total, each child was exposed to a total of 120 word-picture pair trials, half of which were congruent. Picture stimuli, obtained from the Bank of Standardized Stimuli (BOSS; Brodeur, Dionne-Dostie, Montreuil & Lepage, 2010), were square, digital, colour photos (400x400 pixels) set against a white background. According to printed familiarity norms from the MRC Psycholinguistic Database (Wilson, 1988), congruent labels were highly familiar (mean = 548.32; SD = 49.20). They also had high name agreement (mean = 91.4%; range = 81-100%; Brodeur et al., 2010; Brodeur, Guerard & Bouras, 2014). Mean congruent word frequency was 194.43 per million words (range = 5-995), as read in print by children aged 5 to 9 years old (Children's Printed Word Database, 2002).

Each congruent spoken word label was matched with an incongruent spoken word label. The Children's Printed Word Database was used to match incongruent words with congruent words on initial letter sound, syllable length and frequency (p = 0.975), while the MRC Psycholinguistic Database was used to match the words on imageability (p = 0.412), familiarity (p = 0.779) and concreteness (p = 0.107; see Appendix A for full list of congruent and incongruent word-picture stimuli). Words in each condition ranged from one to three syllables in length (mean = 1.42). Picture labels were examined using semantic association norms (Nelson, McEvoy & Schreiber, 1998), in order to confirm that items in each incongruent pair were not meaningfully related to one another.

The order of word-picture pair presentations was pseudorandomised, so that no more than three presentations of either a congruent or incongruent pair occurred consecutively. Trials containing the same picture were separated by at least ten other trials. For half of the trials, pictures appeared for the first time with their congruent word labels, while for the remaining half, pictures appeared for the first time with their incongruent word labels. During the session, the 120 experimental trials were divided into four blocks of 30 trials each, with a small break provided for participants between each block.

4.3.4.2. Auditory stimuli. All spoken word stimuli were recorded using a Rode NT1-A Studio Cardioid Condenser Microphone (frequency range 20Hz to 20 000Hz, sensitivity -31.9dB re 1 V/Pa [25.00mV at 94dB SPL]), mounted on a Rode Desk DS1 Desk Stand with SM6 shock mount with pop filter and connected via a shielded XLR cable to an RME Fireface UC external sound card. This sound card was connected to a laptop personal computer running Audacity for Windows (version 2.0.2) and TotalMix FX for Windows (version 0.998) software. Key parameters set in the TotalMix FX for Windows software for these recordings included a mono recording with a sampling frequency of 44.1kHz, a gain setting of 22.0, a 48-volt phantom power for the condenser microphone, and all equalisers (filters) disabled. All recordings were saved in wave file format at the original 44.1kHz sampling rate with 32-bit resolution.

All spoken word stimuli were recorded by the same adult female native Australian English speaker (author Bell) in a sound-treated audiometric booth (3.2 x 3.2 x 2.25m). Each word was spoken five times, with all utterances recorded in a single session on a single wave file (wave audio file format) at a 44.1kHz sampling rate with 32-bit resolution. Each wave file was edited on a personal computer running Audacity (version 2.1.2). The ‘best’ version of each word was selected and saved as an individual wave file. Two raters (authors Bell and Angwin) independently identified the best recording of each word based on perceptual rating of clarity. The two raters then came together to reach consensus on any items where they disagreed. The final individual wave files were normalised through the normalisation function (written by Dominic Mazzoni) of the Audacity for Windows software (version 2.1.2) to a maximum output level of -1.0dB. This effectively increased the amplitude of each word stimulus to the maximum possible without distortion (peak clipping).

4.3.5. Procedure. Two EEG tasks lasting approximately the same duration were administered to each child: the word-picture matching task and a rhyme judgement task (relating to a separate study; see Chapter 5). To control for the effects of fatigue, the order of task completion was balanced between groups, so that approximately half of each group completed the word-picture task first.

During the EEG task, each participant was seated upright in a comfortable chair at approximately 0.5-1 metre distance in front of two speakers (Altec Lansing Model 220, frequency response 70Hz to 18000Hz) in a sound-treated and radio-frequency-shielded audiometric booth (3.2 x 3.2 x 2.25m). The stimuli were presented through these speakers via a personal computer running E-Prime 2.0. Speaker volume was adjusted for each participant, depending on whether they were in the TH or CI group. Stimuli output levels peaked in the range of 74 to 76 dB SPL for CI children and 54 to 56dB SPL for TH children. These levels were measured with a Bruel and Kjaer 2250 SLM (class 1) with a type 4189 ½ inch free field microphone, set to record on a slow (1s) setting, at 0.8m from the speakers at the approximate height of a participant's head. Words ranged in duration from 0.35 to 0.78 seconds. Visual stimuli for the task were presented on a 34x27cm monitor screen. For the entire duration of the EEG testing, participants were accompanied in the booth by an adult experimenter (author Bell), who recorded verbal responses electronically and manually initiated each trial as appropriate.

Participants were advised that they would hear a spoken word through speakers, and this would be followed by a picture in the centre of their computer screen which sometimes matched the picture and sometimes did not. The children were asked to wait for a response prompt after the picture and to then respond verbally, with 'yes' if the stimuli matched, or 'no' if they did not match. A short practice block was presented prior to the actual experiment, during which the experimenter addressed any observed difficulties relating to participants' understanding of the task requirements. The stimuli used in practice trials were different to those used in the experimental trials, but had similar lexical characteristics (i.e., matched on initial letter sound and syllable length, and no significant difference in frequency, familiarity, concreteness and imageability). The sequence of events for a typical trial was: (1) a fixation cross in the centre of the screen for 500ms; (2) blank screen lasting 2000ms while spoken word is played (all spoken words offsetting at 1000ms); (4) coloured picture in the centre of the screen for 1000ms; (5) a blank screen for a further 500ms, to minimise motor response interfering with EEG recording; (6) a question mark in the centre of the screen to prompt participant's response. This sequence is illustrated in Figure 4.1.

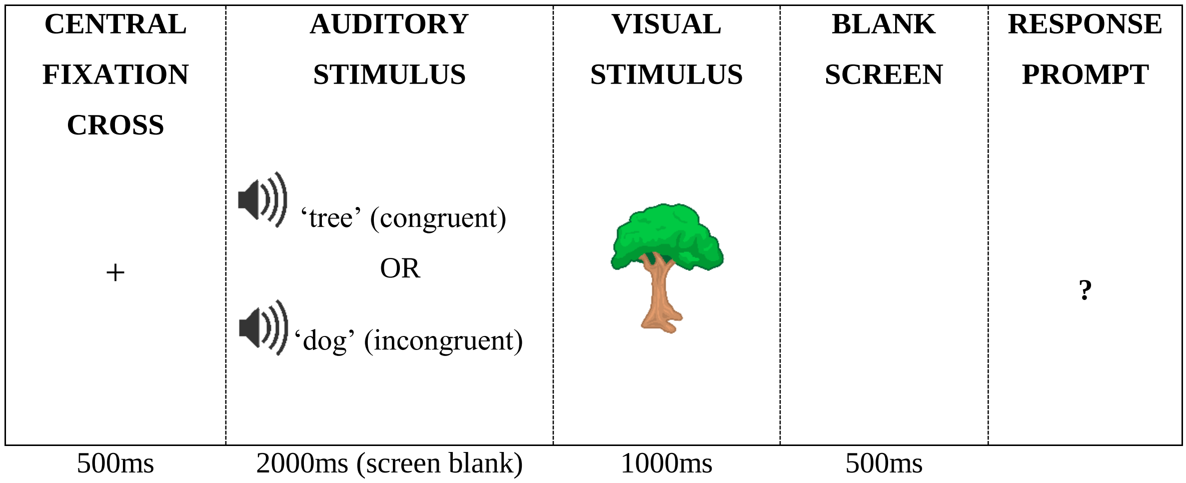

Figure 4.1. Sequence of a typical word-picture matching task trial.

4.3.6. EEG recording and analysis. Event-related potentials were recorded with a 128 channel Electrical Geodesics, Inc. system, with a Net AMPS EEG Amplifier. The Electrical Geodesics, Inc. sensor net contained Ag-AgCl electrodes surrounded by sponges soaked in a saline solution (potassium chloride and water). Eye blinks were monitored using infra- and supra-orbital electrodes and electrodes on the external canthi. EEG recording occurred continuously, with a sampling rate of 500Hz. Impedances were kept below 50kΩ, which is an acceptable level with the use of high impedance amplifiers (Ferree, Luu, Russell & Tucker, 2001).

Analyses were conducted using Net Station software (version 4.5.1) on a MacIntosh computer. After application of high pass (0.1Hz) and low pass filters (30Hz), EEG data pertaining to correct responses were segmented into 900ms epochs (starting 100ms before the target picture onset). For both groups, an automatic artefact detection procedure was used to mark and replace bad channels. A channel was marked as bad for a given segment if any two points within that channel for that segment differed in voltage by at least 200µV. Data from bad channels were replaced with data interpolated from the remaining channels. An automatic artefact detection tool was also used to mark eye blinks. A segment was marked as containing an eye blink if any two points within either the vertical or horizontal electrooculogram channels contained points that differed by at least 140µV. Visual inspection procedures were then employed by the researchers to verify the presence of artefacts for each trial. For the CI group, electrodes in direct contact with the outer components of the CIs were excluded from analyses. Visual inspection confirmed that there were no significant CI artefacts, as is consistent with previous ERP research involving the presentation of visual linguistic stimuli in children with CIs (e.g., Kallioinen et al. 2016).

ERP data for two participants, both from the TH group, were excluded because of excessive EEG artefacts. For each of the remaining 28 TH children and 12 children with CIs, electrode data was re-referenced to an average reference, correcting for the polar average reference effect, then baseline corrected for 100ms prior to this point. The average number of accepted trials for semantically congruent word-picture pairs was 36.95 in children with CIs and 37.61 in children with typical hearing. The average number of accepted trials for the incongruent condition was 38.25 in children with CIs and 36.54 in children with typical hearing.

Six EEG channels, comprising Cz and the five channels directly adjacent to Cz, were included in analyses (see Figure 4.2). This distribution of channels was expected to capture the N400 effect, which is generally broad and encompasses fronto-central, central and centro-parietal regions (Byrne et al., 1999; Henderson et al., 2011). As stated previously, electrical artefacts from CIs were not observed, though in order to further reduce the potential for contamination, it was considered important to restrict statistical analyses to predominantly midline electrodes, which were distal to the outer components of the CIs. This step is consistent with past research involving CI participants (e.g., Hahne et al., 2012). The specific region of interest, according to the 128-electrode GeoDesic sensor net system, included channels 7, 106, 31, 80, 55 and the reference channel. These channels respectively approximate C1, C2, Cp1, Cp2, Cpz and Cz in a 10-10 system. EEG data were averaged across these six electrodes for all analyses.

The mean amplitude of the N400 effect for each participant was quantified by calculating the difference wave between the congruent and incongruent conditions. Each participant's peak latency of the N400 effect was measured as the latency of the most negative voltage value reached for the same difference wave. Mean amplitude and peak latency values were calculated based on the time region of 300 to 500ms post-onset of the picture stimuli, as averaged across the six channels of interest. This time region was selected with reference to the N400 effect latencies reported in prior research with similarly aged cohorts of children (e.g., Cummings & Ceponiene, 2010; Henderson et al., 2011; Kallioinen et al., 2016). Visual inspection of each group's grand-averaged ERP data confirmed that there was a large negative deflection between 300 and 500ms, peaking at a more negative amplitude for the incongruent condition; thus, this time region was considered suitable for analyses. A repeated measures ANOVA was performed to explore the mean amplitude and peak latency results within and between groups.

4.4. Results

4.4.1. Behavioural results. A summary of group comparison results pertaining to measures included in the present study's analyses (n = 40) is provided in Table 4.2. The TH group performed significantly better than the CI group on measures of word-level comprehension (i.e., receptive vocabulary) and sentence-level comprehension, but not on passage-level comprehension. The TH group also obtained significantly higher scores on written language measures of word reading accuracy and passage reading comprehension.

Generally speaking, the accuracy of responses to the ERP word-picture matching task was high. The average proportion of correct responses in the TH group was 94.83% for congruent trials (SD = 4.80%) and 97.11% for incongruent trials (SD = 3.97%). The average proportion of correct responses in the CI group was 93.47% for congruent trials (SD = 4.17%) and 95.42% for incongruent trials (SD = 4.67%). Since the assumption of normality for behavioural accuracy data was violated, a non-parametric approach (i.e., Mann-Whitney U test) was taken to confirm there was no significant difference between groups in the proportion of accurate trials for either congruent ($p = 0.153$) or incongruent ($p = 0.229$) conditions.

4.4.2. N400 effect. To compare the mean amplitude of the N400 effect between TH and CI groups, a repeated measures ANOVA was conducted, using the between-subjects factor of group (TH, CI) and within-subjects factor of condition (congruent, incongruent). A main effect of condition was found, $F(1, 38) = 41.10$, $p < 0.001$, $\eta_p^2 = 0.520$. There were no significant interactions between group and condition, indicating that the TH children and the children with CIs demonstrated a similar N400 effect (Figure 4.2).

In order to quantify the mean amplitude and peak latency of the N400 effect within each group, a difference wave was generated by subtracting the congruent condition waveform from the incongruent condition waveform. Each group's mean amplitude of the N400 difference wave was then entered into an independent samples t-test, which confirmed that there was no significant differences between groups. Each group's peak latency of the

N400 difference wave was also included in an independent samples t-test and again, there was no significant difference between groups.

4.4.3. Relationships between the N400 effect and behavioural measures. Of additional interest in the present study was whether the N400 effect correlated significantly with behavioural measures of word-level oral comprehension, sentence-level oral comprehension, passage-level oral comprehension, word reading accuracy and reading comprehension. These behavioural measures were included in Pearson correlational analyses, along with mean amplitude and peak latency values for the N400 effect, calculated using the difference waves between conditions. The CI and TH groups were examined separately for all correlational analyses.

For the TH group, the correlation between sentence-level language comprehension and amplitude of the N400 effect approached significance, $r = -.36$, $p = 0.060$ (Table 4.3). No other correlations reached or approached statistical significance for the TH group, and there were no significant correlations for the CI group.

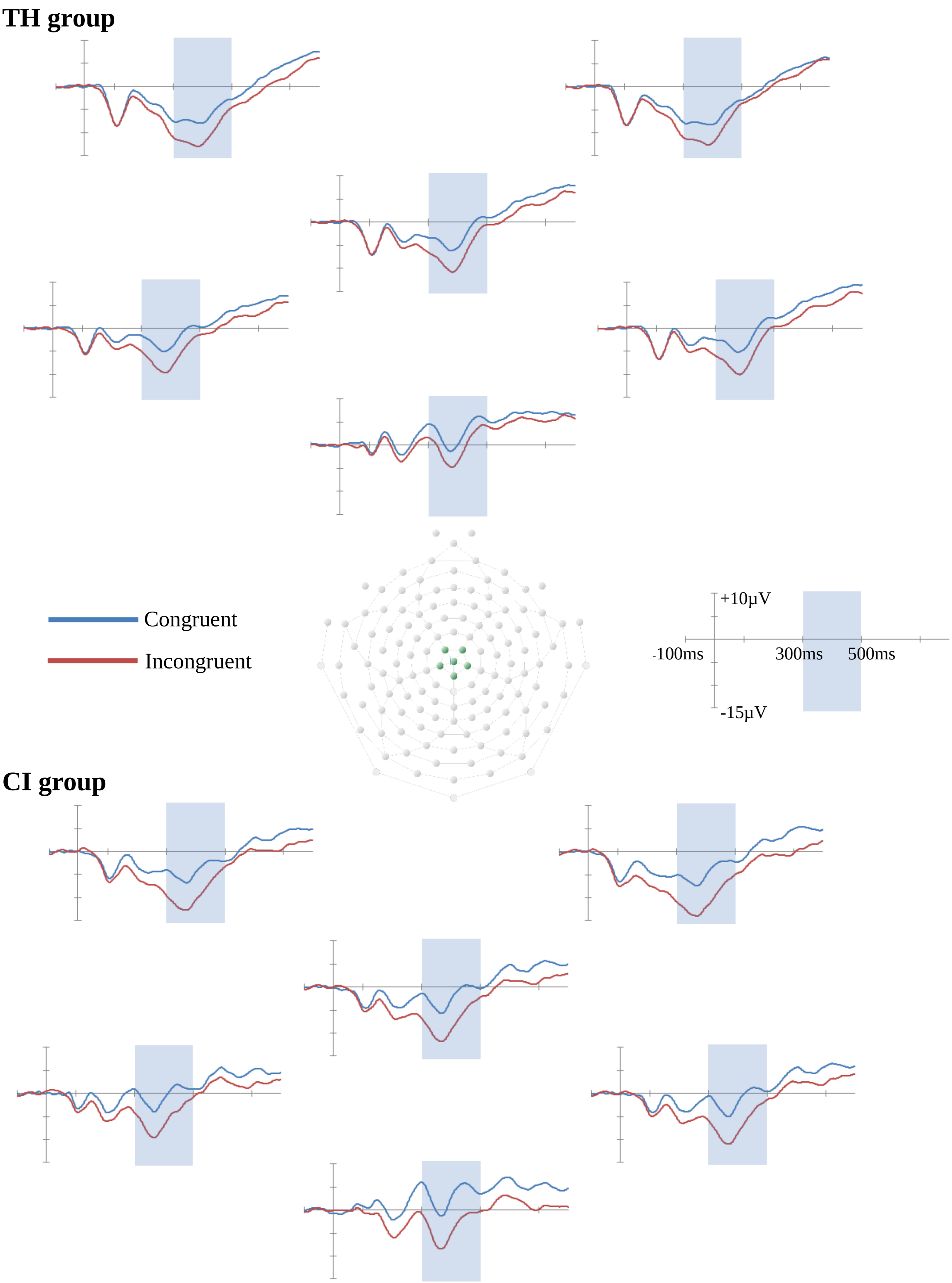

Figure 4.2. Grand-averaged ERPs for congruent and incongruent conditions in each group.
Note. TH = typically hearing; CI = cochlear implant. Shading used to indicate the time period after target onset (i.e., 300-500ms) captured in N400 effect analyses. Six channels within centro-parietal region of interest were used in analyses (highlighted green in channel map).

Table 4.2

CI vs. TH group comparisons on behavioural language and reading measures.

Domain	Measure	CI (n=12) Mean (SD)	TH (n=28) Mean (SD)[1]	*t*	Cohen's *d*	*p*
Written language	Word reading	-0.37 (1.00)	.56 (1.29)	2.220	.81	*.032
	Reading comprehension	96.25 (13.12)	105.82 (13.93)	2.024	.71	*.025
Spoken language	Word-level vocabulary	99.83 (17.66)	116.82 (14.57)	3.170	1.05	*.003
	Sentence-level comprehension	8.42 (2.31)	10.79 (2.54)	2.769	.98	*.009
	Passage-level comprehension	8.75 (2.67)	9.79 (2.44)	1.197	.41	.239

Note. CI = cochlear implant; TH = typically hearing. Statistically significant values (p<.05) indicated with asterisk (*). Standard score (normative mean = 100) reported for word-level comprehension (i.e., receptive vocabulary) and reading comprehension; standard scaled score (normative mean = 10) reported for sentence- and passage-level comprehension; standardised z-score (normative mean = 0.0) reported for word reading.

Table 4.3

Pearson correlations between ERP and behavioural measures (TH [n=28] and CI [n=12] groups).

		Word-level comprehension	Sentence-level comprehension	Passage-level comprehension	Word reading	Reading comprehension
TH N400 effect	Amplitude	-.29	-.36	-.03	-.01	.11
	Latency	-.13	-.19	-.27	-.18	-.19
CI N400 effect	Amplitude	.30	-.17	-.07	.04	-.11
	Latency	.31	-.27	.11	-.05	-.15

Note. ERP = event-related potential; TH = typically hearing; CI = cochlear implant.

4.5. Discussion

The present study sought to determine whether beginning readers with CIs demonstrated a typical sensitivity to semantic incongruence at the level of neurological functioning. Results indicated that children with CIs showed a typical N400 effect when participating in a word-picture matching task: the ERP waveform elicited during the task was significantly more negative when paired stimuli were incongruent (i.e., non-matching), compared to when they were congruent (i.e., matching). Moreover, the amplitude and latency of the N400 effect did not differ to that of TH children of a similar chronological and mental age. No significant correlations between ERP and behavioural measures were found, although the correlation between sentence-level language comprehension and amplitude of the N400 effect approached significance for the TH group. Interpretations pertaining to observed results are described in detail below.

4.5.1. The N400 effect in children with cochlear implants. The prevailing view of the 'N400 effect', when elicited by semantically predictable and unpredictable stimuli (e.g., words), is that it indexes the retrieval and contextual integration of lexical-semantic information (Kotz & Friederici, 2003). The comparable N400 effects elicited in the present study's CI and TH groups suggest that the children with CIs could efficiently process the semantic representations of word stimuli. To the author's knowledge, this is the first report of such a finding in beginning readers with CIs who communicate solely via spoken language.

Supporting the present study's results are similar reports of N400 effects in younger hearing-impaired children. Vavatzanidis and colleagues (2018) found that children aged approximately 2 to 6 years old, who had received bilateral CIs one to two years prior to testing, demonstrated a significant N400 effect in response to congruent and incongruent picture-word pairs. Similarly, in a case study reported by Key and colleagues (2010), a significant N400 effect was elicited in a 6-year-old child with profound bilateral hearing loss, four months after she received her second CI. In contrast, the effect was absent prior to the activation of the second CI, during which time she completed the task having only unilateral auditory stimulation (Key et al., 2010). While not conclusive – especially given the lack of TH control group (Vavatzanidis et al., 2018) and single case sample size (Key et al., 2010) – taken together, the results from these studies provide some context for the early language development of children with significant hearing loss. Cochlear implants allow this population a means of accessing spoken language, which may therefore facilitate the development of aural-based semantic representations.

Challenging the present study's results are differing reports of N400 effects by Kallioinen et al. (2016), albeit in hearing-impaired children who differed from the children in the present study by their younger age (5 to 7 years old) and their use of both sign and spoken language. Children with CIs displayed a significant N400 effect, supporting the assertion that this population can retrieve and integrate word-level semantic information. However, the N400 effect elicited in the study's between-category condition, which was the condition most similar to that in the present study, was larger than the effect elicited in a similarly aged TH control group. Such a finding was unexpected, especially as some scholars have associated larger N400 effects with *better* semantic processing abilities (e.g., Cummings & Ceponiene, 2010), and better semantic processing abilities were not reflected in the CI cohort's response to behavioural tasks with high semantic processing demands (Kallioinen et al., 2016). The authors posited that the presentation of more incongruent stimulus pairs than congruent stimulus pairs may have encouraged their TH group to take a more passive, bottom-up approach to the task, thereby reducing their ERP effects of semantic relatedness.

Kallioinen et al. (2016) also examined the ERP waveforms elicited by more fine-grained semantic judgements wherein incongruent trials were semantically related. The amplitude of the N400 effect for the CI group was significantly smaller – though still present – in response to this 'within-category' condition, relative to the 'between-category' condition. In contrast, the TH children demonstrated similar N400 effect amplitudes across the two conditions. Future research to replicate and extend Kallioinen et al.'s findings should be considered, particularly with respect to exploring how manipulations to the semantic characteristics of stimuli influence ERP responses within the CI population, while controlling for the strategic demands of the EEG task.

Aspects of the present study's experimental paradigm could also be altered in future research to gain a better understanding of how individuals with hearing loss process language. Hahne and colleagues (2012) employed a cloze sentence task with adult CI users, and found that the N400 effect elicited in this group was longer-lasting than that of TH adults. A similar task could be used in children with CIs to provide further insight into their lexical-semantic processing skills in a sentence comprehension context. Along another line of inquiry, it would also be worthwhile to examine the development of semantic representations in individuals with hearing loss who use sign language to communicate. In the present study, all children used only spoken communication. This criterion was imposed in order to reduce the potentially confounding effect of communication mode, which has been found to influence language outcomes in the past (e.g., Cupples et al., 2017; Johnson & Goswami, 2010). As such, a potential limitation is that results do not necessarily generalise to children with hearing loss who do not use spoken communication. Future research may address whether the neural response to a picture-word (or picture-gesture) paradigm is altered in users of sign or total communication, whose lexical-semantic representations develop via exposure to non-auditory information.

Despite the present study's finding that children with CIs who use spoken communication produced an N400 effect that was similar to that of TH children, there were still significant differences found between the groups in terms of performance on behavioural measures of spoken language and reading. On the surface, this may be considered surprising, since prior studies have uncovered differences in the size and shape of ERP waveforms elicited in certain clinical populations. For example, children with language disorders demonstrate comparatively reduced (Cummings & Ceponiene, 2010) or delayed (Pijnacker et al., 2017) N400 effects, and similar findings have also been reported for children with dyslexia (Jednoróg, Marchewka, Tacikowski & Grabowska, 2010; Stelmack & Miles, 1990). However, the cohort with CIs included in the present study differ to those examined previously, since the spoken and written language skills of the present cohort were, on average, within the normal range. Hence, in the context of existing research involving different clinical populations of children, the present study's results indicate that the N400 effect may be insensitive to group differences when the performance of the population of interest on behavioural measures is within normal limits. The question of how strongly a group's ERP response relates to their actual language and reading skills was investigated further, as described in the following section.

4.5.2. Relationships between ERP and behavioural measures. No correlations between ERP and behavioural results were statistically significant in the present study. However, the correlation between sentence-level language comprehension and N400 effect amplitude approached significance in the TH group (r = -0.36, p = 0.060), such that a larger mean amplitude was associated with better comprehension. The potential existence of this relationship is supported by similar findings reported in the ERP literature. Correlations between the N400 effect amplitude and language comprehension have been found in 8- to 10-year-old children (Henderson et al., 2011) and 4- to 6-year-old children with SLI (Pijnacker et al., 2017). Moreover, the amplitude of N400 effect is also reduced in adults with poor spoken language comprehension skills, relative to those with better skills (Landi & Perfetti, 2007). Based on the observed trend in results from past studies, as well as those from the present one, it may be posited that the language skills required for comprehending spoken language are also required for classifying stimuli as congruent or incongruent based on the preceding context.

For children with CIs, no significant correlations were observed between the amplitude or latency of the N400 effect and skills in spoken language and reading. Importantly, a limitation of the present study was the small sample size of the CI cohort, and hence, the observed null findings need to be interpreted with caution. There is also little precedent in the literature on which to base concrete interpretations. Kallioinen and colleagues (2016) also failed to find a significant correlation between the N400 effect and behavioural assessment measures when 'incongruent' items were not from the same semantic category, as was the case with our stimuli. In contrast, when Kallioinen et al. (2016) examined the 'within-category' N400 effect, there was a significant relationship between the N400 effect amplitude and phonological processing, as well as cloze sentence completion. It is possible that more fine-grained judgements about semantic congruence elicit an N400 response that better represents spoken language skills, as assessed by behavioural measures. This interpretation is, however, limited by the fact that Kallioinen et al., (2016) did not assess spoken language comprehension, *per se*. Future research into the accuracy with which the N400 effect represents actual language skill, using more closely related stimuli and a greater sample size of children with CIs, is therefore warranted.

4.5.2.1. *Vocabulary and the N400.* For both groups, no relationship was found between receptive vocabulary and the N400 effect, which was surprising given the similarity in stimulus presentation characteristics and the semantic processing demands of the vocabulary and ERP tasks. Indeed, more complex sentence-level comprehension skills appeared to be better related to ERP results for the TH group. The non-significant correlations between vocabulary and the N400 effect amplitude are not considered a spurious finding when viewed in the context of past literature. Those studies involving the paired presentation of congruent and incongruent stimuli also report weaker relationships between N400 effects and word-level measures, when compared to broader language comprehension measures (Henderson et al., 2011; Landi & Perfetti, 2007; Pijnacker et al., 2017).

An interesting distinction can be drawn between our own study and those that have elicited the N400 component using written words in isolation, rather than words preceded by a congruent or incongruent stimulus. Such investigations have, in fact, uncovered significant relationships between the amplitude of the N400 component and vocabulary (Coch & Benoit, 2015; Khalifian, Stites & Laszlo, 2015; Stites & Laszlo, 2017). According to Stites and Laszlo (2017), the N400 component observed in their passive word exposure task likely indexed the neural processing associated with semantic access, and hence it was reasonable to expect that the size of an individual's vocabulary influenced the amplitude of this response. In contrast, the present study's paradigm involved the presentation of a target item, subsequent to the presentation of another item. Hence, rather than eliciting a search for existing vocabulary-based information, exposure to the target may have elicited a response that was associated with the integration of an item with its context, such that the individual could classify it as congruent or incongruent. The N400 effect has previously been elicited by manipulating the lexical-contextual integration demands associated with processing sentences (Laszlo & Federmeier, 2009). It is these integration skills, rather than single item-based vocabulary skills, that may have been indexed by the N400 effect in the present study, which may account for the trending relationship between N400 effect amplitude and sentence-level comprehension in TH children.

The aim of the above discussion is not to argue that language comprehension at a broad level is influenced solely by the lexical-semantic integration skills of an individual, as indexed by a specific neural response that occurs within half a second of exposure to linguistic stimuli. Certainly, there appears to be a clear dissociation between the precise processes measured by behavioural and ERP testing approaches, based on the weak and non-significant correlations reported in the present study, as well as in previous research (e.g., Coch & Benoit, 2015; Kallioinen et al., 2016; Pijnacker et al., 2017). Hence, while not directly applicable to a clinical or educational setting, ERP experiments nevertheless help to provide a more complete picture of what language processing 'looks like'. Skills in spoken and written language develop on the basis of many underlying processes, and a coherent understanding of the true complexity of this development can only arise from investigations that focus on these processes in fine detail, as has been the aim of the present study.

4.6. Conclusion

This study used a word-picture matching paradigm to examine the semantic processing of school-age children with CIs. A significant N400 effect was elicited in the CI group, and the characteristics of this effect were comparable to a TH control group. At a neural level, children with CIs appeared able to integrate lexical-semantic information in a normal manner, although their behavioural spoken and written language performances were still significantly below the level of their peers. The dissociation between ERP and behavioural measures was also highlighted by an absence of significant correlations between language measures and N400 effect characteristics in both CI and TH groups. That said, the correlation between sentence-level comprehension and amplitude of the N400 effect approached significance for TH children, and this was posited to represent the shared lexical-semantic integration demands of spoken language processing and word-picture matching. As one of only a few studies to use an ERP methodology to investigate linguistic processing in children with CIs, the results presented here provide the impetus for future studies to replicate the findings with a greater sample size. Further research may also explore whether group differences between cohorts with and without hearing loss emerge when ERP stimuli are systematically manipulated for semantic relatedness (e.g., within-category incongruence versus between-category incongruence) or even modality (e.g., picture-gesture matching).

Chapter 5.

Phonological Processing in Children with Cochlear Implants: ERP Evidence for the Rhyme Effect during a Letter Rhyme Judgement Task

As with the content from the previous chapter, the study presented in Chapter 5 aims to provide insight into the neuro-linguistic processing abilities of children with cochlear implants. Here though, the focus moves from *semantic* processing to *phonological* processing, and more specifically, to participants' sensitivity to rhyme incongruence. The results from this chapter also complement the behavioural assessment analyses from Chapters 2 and 3, wherein children with cochlear implants demonstrated difficulties, relative to their typically hearing peers, performing tasks with high phonological demands. In Chapter 5, electroencephalography (EEG) is used to capture children's neural processing of phonological information. Given the fundamental nature of phonological processing in early reading and spelling achievement, the results have important implications for our understanding of literacy development in children with cochlear implants.

5.1. Abstract

The present study sought to explore whether beginning readers with cochlear implants (CIs) demonstrated sensitivity to phonological structure, as indexed by event-related potential (ERP) evidence of a rhyme effect. The participants, aged 6-9 years, responded to a letter rhyme judgement task while their ERPs were recorded. Results pertaining to children with CIs (n = 6) were individually examined, with reference to a control group of similarly aged typically hearing (TH) children (n = 20). Correlations between the rhyme effect and behavioural measures of reading and phonological processing were also analysed in the TH group. According to the findings, a significant rhyme effect was observed in the TH group and, based on case-control analyses and visual inspection, in some of the participants with CIs. There were no obvious links between behavioural and ERP results in the CI cohort, although in TH children, a smaller rhyme effect amplitude was significantly correlated with better letter-sound knowledge and nonverbal reasoning. A high proportion of the original participant cohort were unable to complete the letter rhyme judgement task, suggesting that a certain threshold of cognitive-linguistic skill was necessary to process the task's conceptual demands. On a neurophysiological level, participants' ERP responses may also have been negatively influenced by the ease with which they unintentionally retrieved the target item's 'competing' letter-sound information.

5.2. Introduction

Within an alphabetic orthography such as English, successful early word decoding relies on the young reader's knowledge of how letter sequences represent phonemes (McBride-Chang, 1999). Literacy development therefore takes place on the basis of – and in parallel with – the ability to store, access and manipulate phonological information. Phonological processing deficits are usually presumed to underlie word-level reading disorders in children and adults with typical hearing (Hulme, Snowling, Caravolas & Carroll, 2005; Wagner, Torgesen & Rashotte, 1994). The same hypothesis has also been applied to those with significant hearing loss (Mayer & Trezek, 2014), based on evidence from behavioural assessments wherein these individuals demonstrate difficulty, relative to a typically hearing cohort, performing tasks with high phonological processing demands (Ambrose, Fey & Eisenberg, 2012; Nelson & Crumpton, 2015; Nittrouer, Caldwell, Lowenstein, Tarr & Holloman, 2012; Spencer & Tomblin, 2009). Behavioural assessment results represent the net contribution of a great many underlying neural processes (Henderson, Baseler, Clarke, Watson & Snowling, 2011). Hence, research conducted using behavioural assessment methods may be expanded upon by neurophysiological research, of which there is none to date involving children with any degree of hearing loss. The present preliminary study examined how beginning readers with severe-to-profound hearing loss, who have cochlear implants and use spoken communication, process rhyme, as evidenced by their neural response to an electroencephalography (EEG) task.

Past studies have examined the processes underlying linguistic development by combining EEG technology with experimental paradigms to elicit predictable patterns of neural activity. These patterns, known as 'event-related potentials' (ERPs), are presumed to index significant changes in post-synaptic activity, time-locked to occur following exposure to a certain stimulus (Kutas, Van Petten & Kluender, 2003). Different ERP components are extracted from EEG activity, as characterised by the number and polarity of ERP peaks, the latency with which these peaks occur, the distribution of activity across the scalp, and the exact stimulus to which the ERP is generated in response (Kotz & Friederici, 2003). The precise temporal resolution of ERP measurement makes it a useful tool for studying language processing, which is based on the functioning and coordination of many small-scale mechanisms. In this way, ERP data can complement data obtained via more traditional testing methods.

With regard to phonological processing in particular, past studies have examined an electrophysiological phenomenon referred to as the ‘rhyme effect’, or the ‘N450’ (e.g., Coch, Mitra, George & Berger, 2011; Grossi, Coch, Coffey, Corina, Holcomb & Neville, 2001; MacSweeney, Goswami & Neville, 2013). The rhyme effect represents the difference in neural response elicited by rhyming versus non-rhyming stimuli. Specifically, the amplitude of the ERP waveform, between 300 and 600 milliseconds post-onset of a target stimulus (e.g., ‘hat’), is less negative when preceded by a rhyming prime (e.g., ‘cat’), compared to a non-rhyming prime (e.g. ‘tree’; Grossi et al., 2001). Parallels may be drawn between the rhyme effect and the semantically driven ‘N400 effect’, given that both represent the neural response to a stimulus that is incongruent with the preceding context (Kutas & Federmeier, 2011). However, as the rhyme effect has been elicited using non-lexical items, such as nonsense words (Coch, Hart & Mitra, 2008) and letters (Bann & Herdman, 2013), it is thought to index phonological sensitivity rather than semantic processing (Bann & Herdman, 2013).

Rhyme awareness, as measured by behavioural testing methods, is considered one of the earliest indicators of phonological sensitivity (Ziegler & Goswami, 2005), and it has also been found to significantly predict school-age literacy development (de Jong & van der Leij, 2003). Accordingly, it is unsurprising that existing ERP research has focused on eliciting the rhyme effect in children with reading difficulties. A study by Ackerman, Dykman and Oglesby (1994) revealed abnormal rhyme effects in 7- to 12-year-old children with dyslexia. When compared with groups of age-matched ‘slow learners’ and children with attention-deficit disorder, the peak amplitude of the effect elicited by a real and nonsense word visual rhyme judgement task was significantly reduced in the dyslexic group. Similarly, two other studies have reported attenuated rhyme effects in adolescents with dyslexia, although interestingly, this attenuation was restricted only to those with ‘dysphonetic’ dyslexia, whose nonword reading scores were poor (McPherson, Ackerman, Holcomb & Dykman, 1998; McPherson, Ackerman, Oglesby & Dykman, 1996). These findings provide support for the rhyme effect indexing skills in phonological processing.

That said, attenuated rhyme effects have not been universally observed in individuals with reading difficulties. Noordenbos, Segers, Wagensveld and Verhoeven (2013) used an auditory word rhyme judgement task to investigate the ERP responses of 6- and 7-year-old children at genetic risk of dyslexia. This cohort demonstrated a significant difference between rhyming (e.g., wall-ball) and non-rhyming (e.g., sock-ball) conditions, and the amplitude and latency of this response was comparable to an age-matched control group. As in prior studies (e.g., Ackerman et al., 1994), Noordenbos et al's 'at-risk' group demonstrated significantly worse real and nonword reading skills than their typically developing peers. Hence, it is not immediately clear what accounts for the comparatively normal rhyme effects reported in this group but not elsewhere (e.g., Ackerman et al., 1994).

One factor to consider in evaluating the results of past studies, including that by Noordenbos and colleagues (2013), is the use of real word stimuli, and thus the introduction of potentially confounding lexical-semantic processing demands. Abundant research has investigated the previously referenced 'N400 effect', wherein the semantic relatedness of two items, presented consecutively, modulates the amplitude of the waveform approximately 400 milliseconds after presentation of the second item (Kutas & Federmeier, 2011). The use of real word pairs in a task that measures *phonological* incongruence therefore makes the interpretation of results challenging (Coch, Grossi, Skendzel & Neville, 2005), especially if lexical-semantic factors have not been controlled across conditions, as appears to be the case in the aforementioned studies (Ackerman et al., 1994; McPherson et al., 1996; 1998; Noordenbos et al., 2013).

The degree of orthographic relatedness between prime and target letter strings has also been found to modulate ERP responses, such that the presentation of a target item elicits a different waveform when preceded by an orthographically similar item (e.g., horle-horse), compared to when it is preceded by an orthographically dissimilar item (e.g., hiele-horse; Savill & Thierry, 2011). This effect may be associated with the finding that, more broadly, exposure to a real or nonsense word activates semantic information associated with the item's orthographic 'neighbours' (Laszlo & Federmeier, 2011). Accordingly, analyses of the rhyme effect may be confounded if rhyming stimulus pairs (e.g., flower-power) are more orthographically similar than non-rhyming stimulus pairs (e.g., build-power), since semantic information pertaining to the target item will be partially activated from exposure to the prime. In order to limit the influence of orthographic similarity on observed rhyme effects, many studies have only included dissimilar stimulus pairs (e.g., flower-hour; Grossi et al., 2001). Some have also included single letters as paired stimuli, thus removing the potential for word-level orthographic and semantic confounds entirely (Coch et al., 2011).

Using letter pairs that rhyme (e.g., A-J) and do not rhyme (e.g., A-G), the rhyme effect has been examined in a number of studies involving typically hearing adults (Bann & Herdman, 2016; Coch, George & Berger, 2008; Coch, Hart & Mitra, 2008). The robustness of the rhyme effect within such studies is not affected by visual characteristics such as letter case (Coch, George & Berger, 2008). Moreover, the effect has been elicited in paradigms where participants were not required to make overt rhyme judgements about letter stimuli (Stevens, McIlraith, Rusk, Niermayer & Waller, 2013). Hence, the effect of rhyming condition observed in a letter rhyme paradigm is thought to index the processing of phonological information, rather than the processing of physical orthographic properties (Coch, George & Berger, 2008) or judgements about rhyming status (Stevens et al., 2013).

The letter rhyme paradigm has also been used to investigate rhyme effects in children. Coch and colleagues (2011) found a significant rhyme effect in 6- to 9-year-old typically hearing beginning readers. The effect was comparable in latency and mean amplitude to a similarly significant rhyme effect elicited in a separate group of adults, indicating that the neural systems underlying task performance appear well established from a young age (Coch et al., 2011). No significant correlations were found between the amplitude or latency of the rhyme effect and behavioural measures of letter name knowledge, word reading, nonword reading, phonological awareness or phonological memory. This finding suggests that there is some dissociation between the phonological processes that are measured on-line via electrophysiological recording, and those that are measured offline in untimed behavioural assessments (Coch et al., 2011). Importantly, letter-sound knowledge was not among the behavioural measures included by Coch et al. (2011), although such knowledge could theoretically influence the neural activity elicited in response to letter stimuli, particularly for beginning readers receiving phonics education at school. To investigate the relationship between the rhyme effect and letter-sound knowledge, this behavioural measure was included in the present study.

5.2.1. The rhyme effect in populations with hearing loss. Electrophysiological research into the phonological processing abilities of adults with hearing loss is, in its current state, sparse and conflicting. MacSweeney et al. (2013) used a visual rhyme judgement task with paired word stimuli to elicit the rhyme effect in adult sign language users with severe-to-profound congenital hearing loss. The cohort, of whom there were nine participants with useable ERP data, demonstrated a significant effect of rhyming condition, and the amplitude and latency of this response was comparable to a group of typically hearing adults matched on sex, age, nonverbal IQ and education level. In contrast with these results, Colin, Zuinen, Bayard and Leybaert (2013) did not find a significant rhyme effect in a group of ten adult sign language users, who had hearing loss ranging from moderate to profound. Here, the participants were exposed to a rhyme judgement task with paired picture stimuli. The conflicting results from these two studies make it unclear whether adults with significant hearing loss demonstrate ‘normal’ phonological processing abilities, as evidenced by a rhyme effect.

It is not obvious what accounts for the equivocal findings reported by MacSweeney et al. (2013) and Colin et al. (2013). Possibly, the null results obtained by Colin and colleagues (2013) are associated with the type of stimuli used (i.e., pictures, rather than words); however, given that a typically hearing cohort from the same study *did* produce a rhyme effect, this factor cannot explain the results entirely. In both studies, stimuli were matched between conditions in terms of semantic concreteness, imageability and orthographic similarity, which means the results are also not likely to have been overly influenced by lexical-semantic confounds. The two cohorts are similar with regard to communication mode and age at deafness, which similarly suggests these were not strong factors, although neither study specifies the hearing technology used by participants and this variable may have had some influence on the results. In sum then, further research is needed to tease apart the other potential variables contributing to elicitation of the rhyme effect in adults with significant hearing loss.

To the author’s knowledge, no studies have examined the rhyme effect in children with any degree of hearing loss, despite abundant behavioural evidence for their having phonological processing deficits (e.g., Nelson & Crumpton, 2015). A substantial proportion of those with severe-to-profound hearing loss can access spoken communication via cochlear implants (CIs). However, current hearing technologies do not replicate typical hearing exactly, and it is presumably for this reason that children with CIs are still found to perform worse than their peers on measures of phonological processing (Ambrose et al., 2012; Nittrouer et al., 2012; Spencer & Tomblin, 2009). In order to complement existing behavioural evidence, while simultaneously avoiding the potentially confounding effects of behavioural task demands, the present study examined the rhyme effect in children with CIs, and thus aimed to provide insight into the phonological processing abilities of this population.

5.2.2. Current study. An important benefit of using the letter rhyme paradigm to examine the rhyme effect in an ERP testing context is that word-level orthographic and semantic confounds cannot influence an individual’s neural response to task stimuli. The paradigm may therefore be considered ideal for use with beginning readers, especially where children with reading difficulties are the focus of investigation, as is the case in research involving children with hearing loss. To the author's knowledge, only one study has used the letter rhyme judgement task to elicit a rhyme effect in typically developing young readers (Coch et al., 2011), and no studies have used an ERP paradigm to examine the rhyme effect in children with CIs. To address these gaps in evidence, the first research question in the present study asked: Is phonological processing, as indexed by the mean amplitude and peak latency of the rhyme effect, altered in beginning readers with CIs who communicate via spoken language?

Additionally, this study sought to address the question: Are behavioural reading and phonological processing outcomes related to the amplitude or latency of the rhyme effect, and does the strength of this relationship differ in children with CIs relative to those with typical hearing? In particular, the heretofore unexplored relationship between the rhyme effect (as measured by ERP testing) and letter-sound knowledge (as measured by behavioural testing) was considered important to include in analyses, since an individual's retrieval of a target letter's name will potentially be influenced by their concomitant retrieval of the target letter's sound.

5.3. Method

5.3.1. Participants. Twelve children with CIs and 29 typically hearing (TH) children initially participated in the present study. Most children with CIs and many of the typically hearing participants were recruited from Hear and Say, an organisation that provides audiological and spoken language intervention to children with hearing loss, as well as conducting regular school-based hearing screening. The remaining participants were recruited from the wider community via newsletter advertisements and word of mouth.

All participants adhered to the following inclusion criteria: (1) use of spoken English as native and primary form of communication; (2) nonverbal reasoning at or above normal limits (as measured using the *Raven's Coloured Progressive Matrices*; Cotton et al., 2005; Raven, Raven & Court, 2004); (3) no diagnosed developmental disorders or intellectual disabilities, and; (4) in Grade 1, 2 or 3 of a mainstream school. The CI and TH groups did not differ on gender, $\chi^2(1) = 0.010$, $p = 0.920$; handedness, $\chi^2(1) = 1.459$, $p = 0.227$; age, $t(39) = -1.442$, $p = 0.157$; grade, $\chi^2(2) = 1.471$, $p = 0.479$, or nonverbal reasoning, $t(39) = 0.930$, $p = 0.358$. With regard to nonverbal reasoning, 'normal limits' was operationally defined as performance within one standard deviation of the mean (i.e., a z-score on the *Raven's*

Coloured Progressive Matrices of between -1 and +1). Age and nonverbal reasoning scores were checked to confirm that data were normally distributed within each group and variances were equal between groups. There were no outliers for age or nonverbal reasoning in either group.

Some children from the original (n = 41) participant cohort were excluded from analyses, as they were unable to (1) complete practice trials with sufficient accuracy; (2) complete the first quarter of the task proper with sufficient accuracy, or; (3) complete the first quarter of the task proper with sufficient speed (see Section 5.3.5). As a result of implementing these discontinuation rules, 10 participants (CI = 4; TH = 6) were excluded during the task's administration. The remaining 31 participants (CI = 8; TH = 23) from the original cohort completed the entire letter rhyme judgement task.

Eight participants with CIs (2 left-handed, 3 females) completed the letter rhyme judgement task. Before entering formal schooling, these participants received auditory-verbal therapy (AVT), which was administered from an early age by AVT-certified speech pathologists or Teachers of the Deaf. The mean age at enrolment in early intervention was 10 months (median = 0.29y; range = 0.08-3.25y). The mean age of the CI group at the time of testing was 8.13 years (SD = 0.99y). Participants with CIs were in Grade 1 (n = 1), Grade 2 (n = 3), or Grade 3 (n = 4). The CI group's average nonverbal reasoning z-score on the *Raven's Coloured Progressive Matrices* was 0.92 (SD = 1.37). All participants in the group had bilateral sensorineural hearing loss, and they received CI surgery before the age of 5 years (mean = 2.26y; SD = 1.56y). The mean duration of CI use at the point of EEG testing was 6.04 years (SD = 1.54y). Participants were reported to use their CIs during all waking hours.

Twenty-three participants with typical hearing (3 left-handed, 11 females) completed the letter rhyme judgement task. Children in this TH group passed hearing screening assessments (thresholds of 25dB HL or better at octave intervals of 500 to 4000 Hz) conducted using a commercially available screening audiometer. The participants completed these screenings after entering formal schooling and no more than two years prior to their participation in the present study. The mean age of the TH group was 7.82 years (SD = 0.81y). Two participants were in Grade 1, 12 were in Grade 2, and 9 were in Grade 3 at the time of testing. The group's average nonverbal reasoning z-score on the *Raven's Coloured Progressive Matrices* was 1.18 (SD = 0.94).

5.3.2. Ethics statement. Ethical approval for the present study was obtained from the Behavioural and Social Sciences Ethical Review Committee at the University of Queensland. Gatekeeper ethical approval was also obtained from the Hear and Say Research and Ethical Advisory Committee. All parents gave written informed consent for their children to participate in the study.

5.3.3. Behavioural testing. Behavioural measures were administered by a qualified speech pathologist prior to EEG testing and usually on a separate day (as part of a larger behavioural assessment battery, reported in Chapters 2 and 3). The behavioural assessment for letter-sound knowledge was never administered on the same day as EEG testing, in order to reduce the potential for confusing ERP task demands. Where any behavioural tasks were completed on the same day as EEG testing, their duration did not exceed 10 minutes, and participants were provided with a break before beginning EEG testing.

Real and nonsense word reading accuracy was assessed using the *Castles and Coltheart 2* (*CC2;* Castles et al., 2009*)*, a standardised test normed for Australian children in Grades 1 through 6 (i.e., age 6;0 through 11;6 years). Examinees are asked to read aloud three different word types: (1) nonwords, which have regular spellings but are not real words (e.g., ‘gop’); (2) irregular words, which have irregular English phoneme-grapheme correspondences (e.g., ‘good’), and; (3) regular words, which have regular English phoneme-grapheme correspondences (e.g., ‘bed’). In total, there are 120 items of increasing difficulty (40 of each word type), and each item is presented on an individual card. The examinee is scored 1 for an item if their response accurately reflects the standard adult pronunciation. A score out of 40 for each word type was obtained and this was converted to a standardised z-score, using the test norms. Nonword reading accuracy was represented in the present study by each participant’s nonword standard score. Real word reading accuracy was represented by a composite score, calculated as the average of each participant’s regular and irregular word reading standard scores.

Three subtests from the *Comprehensive Test of Phonological Processing – 2nd edition* (*CTOPP-2*; Wagner, Torgesen, Rashotte & Pearson, 2013) were administered to assess phonological awareness. The CTOPP-2 is a norm-referenced test, with standardised scores available for children aged 4 and over. An index score representing phonological awareness skill was calculated on the basis of children’s performance on: (1) *Elision*; (2) *Blending Words*; (3) *Sound Matching* (4-7 years), and; (4) *Phoneme Isolation* (7+ years). As outlined in the test manual, raw scores from each of these subtests were combined and converted to a composite phonological awareness score. The CTOPP-2 subtests designed to assess symbolic rapid automatised naming were also administered. Rapid automatised naming is defined by the test authors as a measure of phonological retrieval. Examinees are asked to quickly name symbolic items (e.g., digits or letters) in an array. It is not a test of accuracy, but of the speed or efficiency with which the stored label for each item is accessed and pronounced.

Phonological memory was assessed using the *Children's Test of Nonword Repetition* (*CNRep*; Gathercole & Baddeley, 1996). Standardised scores and percentile ranks are available for children aged 4;0 to 9;11 years. In this test, children are asked to repeat a given nonword. There are a total of 40 items on the test, divided equally into two-, three-, four- and five-syllable nonwords. Nonword repetition has been found to reflect phonological processing skills, and is a significant predictor of reading ability in children (Wagner, Torgesen & Rashotte, 1994).

Letter-sound awareness is also an important precursor to reading development (Catts, Herrera, Nielson & Bridges, 2015). The *Letter-Sound Test* (*LeST*; Larsen, Kohnen, McArthur & Nickels, 2011) was used to measure children's explicit knowledge of letter-phoneme correspondences. Australian norms for children aged 5;0 to 9;11 years were used to convert raw scores to standardised z-scores (Larsen, Kohnen, Nickels & McArthur, 2015). In this task, examinees are visually presented with 51 individual letters (or letter groups like 'ch') and are asked to say aloud the associated sound.

5.3.4. ERP testing.

5.3.4.1. Letter stimuli. Based on a similar letter rhyme judgement paradigm used by Coch et al. (2011), experimental stimuli for ERP testing consisted of the uppercase letters A, B, C, D, G, I, J, K, P, T and Y, presented in standard, 56-point size, Arial font. Stimuli were grouped into 64 rhyming and 64 non-rhyming letter pairs. There were 32 different non-rhyming letter pairs, each presented twice over the course of the experiment. There were 24 different rhyming pairs: 16 of these were presented twice over the course of the experiment and eight were presented four times. As per the presentation constraints below, there was an equal number of presentations of each letter in rhyming and non-rhyming conditions. Each participant was exposed to 128 letter pairs in total.

The order of letter pair presentation was pseudorandomised within two lists of 64 pairs. In accordance with the procedures outlined by Coch et al. (2011), pseudorandomisation carried the following constraints: no more than three presentations of either a rhyming or non-rhyming pair occurred consecutively; identical pairs were separated by at least three other letter pair items; prime and target letters were always different to one another (i.e., there was no identity priming), and; no repetition of either the target or prime letter in a stimulus pair occurred over two consecutive trials. In addition, each letter served as both a rhyming and non-rhyming target on an equal number of presentations, and each letter served as the prime of a rhyming pair and the prime of a non-rhyming pair an equal number of times. Stimuli were presented in four blocks of 32 letter pairs (see Appendix B).

5.3.5. Procedure. Two EEG tasks lasting approximately the same duration were administered to each child: the letter rhyme judgement task and a word-picture matching task (relating to a separate study; see Chapter 4). To control for the effects of fatigue, the order of task completion was balanced between groups, so that approximately half of each group completed the letter rhyme judgement task first. Before the letter rhyme task began, participants completed a brief letter naming test, in which they were presented with individual written letters and were asked to provide each letter's name. This task was included to ensure that all children had sufficient letter name knowledge to participate, and at this point, the rhyming or non-rhyming status of letters was not mentioned. The test items included the same stimuli (in the same font and typeface) as were used in the rhyme judgement task. No children were observed to have any difficulty with naming the letters, although initially some children mistook the upper case 'I' for a lower case 'L'. On such occasions, the participant was corrected, and a revised response elicited.

During the EEG task, each participant was seated upright in a comfortable chair at approximately 0.5m distance in front of a monitor screen, in a sound-treated and radio-frequency-shielded audiometric booth (3.2 x 3.2 x 2.25m). Letter stimuli were presented on the 34 x 27cm screen, using a personal computer running E-Prime 2.0. For the entire duration of the EEG testing, participants were accompanied in the booth by an adult experimenter (author Bell), who recorded verbal responses electronically and manually initiated each trial as appropriate.

The rhyme judgement task was first introduced with a description of what it means for two words to rhyme. Examples of rhyming word pairs were also given by the examiner and elicited by participants. Participants were then informed about what to expect in the task. Two letters would be presented consecutively in the centre of the computer screen. Sometimes the two letter names would rhyme with one another, and sometimes they would not. The children were asked to respond verbally to the target stimulus with 'yes' if it rhymed with the preceding letter, and ‘no’ if it did not rhyme with the preceding letter. As per the procedures outlined in Coch et al. (2011), the sequence of events was as follows: (1) a fixation cross (+) in the centre of the screen for 1000 ms; (2) the prime (first) letter in the centre of the screen for 600 ms; (3) a blank screen lasting 1000 ms; (4) the target (second) letter for 600 ms; (5) a blank screen for a further 1000 ms, to minimise motor response interfering with EEG recording; (6) a question mark in the centre of the screen to prompt participant’s verbal response. The sequence for a typical experimental trial is illustrated in Figure 5.1.

FIXATION CROSS	**PRIME**	**ISI**	**TARGET**	**BLANK SCREEN**	**PARTICIPANT RESPONSE**
+	A		K		?
1000ms	600ms	1000ms	600ms	1000ms	

Figure 5.1. Sequence of a typical letter rhyme judgement task trial.
Note. ISI = Inter-stimulus interval.

Two practice blocks, each consisting of the same 6 trials (3 rhyming, 3 non-rhyming) were administered prior to starting the task proper. Following each practice trial, participants received feedback about the accuracy of their response. Explanations and verbal prompts were provided by the examiner during the first practice block, and any observed difficulties or questions associated with the task were addressed at this time. Testing was discontinued if participants scored less than 5 out of 6 in the second block, as this was considered predictive of poor task performance. When actual testing began, the ERP task was discontinued after the first block of letter pair trials (i.e., one quarter of the way through the entire task) if the participant’s accuracy was at or below chance levels (i.e., </=50%), or if the block’s duration exceeded 7 minutes. Two participants (CI = 1; TH = 1) were excluded from testing after the practice trials. Eight participants (CI = 3; TH = 5) were excluded from testing after the first block of task trials. It may be noted that, of the 10 participants who were excluded due to non-completion, this total comprised 6 participants who completed the letter rhyme judgement task first (before the word-picture task), and 4 participants who completed the

letter rhyme task second (after the word-picture task). The order of tasks therefore did not appear to play a strong part in children's completion of the letter rhyme judgement task.

5.3.6. EEG recording and analysis. Event-related potentials were recorded with a 128-channel Electrical Geodesics, Inc. (2006) system, with a Net AMPS EEG Amplifier. The Electrical Geodesics, Inc. sensor net contained Ag-AgCl electrodes surrounded by sponges soaked in a saline solution (potassium chloride and water). Eye blinks were monitored using infra- and supra-orbital electrodes and electrodes on the external canthi. EEG recording occurred continuously, with a sampling rate of 500Hz. Impedances were kept below 50kΩ, which is an acceptable level with the use of high impedance amplifiers (Ferree, Luu, Russell & Tucker, 2001).

Analyses were conducted using Net Station software (version 4.5.1) on a MacIntosh computer. After application of high pass (0.1Hz) and low pass filters (30Hz), EEG data pertaining to correct responses were segmented into 900ms epochs (starting 100ms before the target letter onset). For both groups, an automatic artefact detection procedure was used to mark and replace bad channels. A channel was marked as bad for a given segment if any two points within that channel for the segment differed in voltage by at least 200µV. Data from bad channels were replaced with data interpolated from the remaining channels. An automatic artefact detection tool was also used to mark eye blinks. A segment was marked as containing an eye blink if any two points within either the vertical or horizontal electrooculogram channels contained points that differed by at least 140µV. Visual inspection procedures were then employed by the researchers to verify the presence of artefacts for each trial. For the CI group, electrodes in direct contact with the outer components of the CIs were excluded from analyses. Visual inspection confirmed that there were no significant CI artefacts, as is consistent with previous ERP research involving the presentation of visual linguistic stimuli in children with CIs (e.g., Kallioinen et al. 2016).

A minimum of 70% accuracy, or 90 correct trials, was required for each participant's data to be included in analyses. Of the 31 participants who completed the ERP task, data for four children (two from each group) were excluded because they failed to meet this cut-off. An additional TH participant was excluded because of excessive EEG artefacts. In total then, analyses were conducted with 20 TH children and 6 children with CIs (see Table 5.1 for CI participant information).

Table 5.1

Audiometric information for participants with CIs (n=6).

#	Configuration	Aetiology	Stable (Y/N)	Age (m) at 1st implant	Age (m) at EI enrolment	Unaided PTA (dB)		Aided PTA (dB)	
						Left	Right	Left	Right
1	CI+HA	Con. (genetic-Pendred Syndrome)	N	47	1	96.25	75.00	28.75	*
2	CI+CI	Con. (genetic-Connexin 26)	Y	7	1	≥100	≥100	22.50	18.75
3	CI+CI	Acq. (1;3y; idiopathic)	Y	19	31	≥100	≥100	27.50	22.50
4	HA+CI	Acq. (1;3y; idiopathic; LVAS)	N	50	39	61.25	77.50	30.00	30.00
5	CI+CI	Con. (idiopathic; LVAS)	Y	9	4	76.25	97.50	21.25	22.50
6	CI+CI	Con. (CMV)	Y	8	4	≥100	98.75	25.00	21.25

Note. CI = cochlear implant; HA = hearing aid; EI = early intervention; PTA = pure tone average; Con. = congenital; Acq. = acquired; LVAS = Large Vestibular Aqueduct Syndrome; CMV = Cytomegalovirus. *Data for left ear missing but reportedly similar to right ear.

Electrode data from the final cohort were re-referenced to an average reference, correcting for the polar average reference effect, then baseline corrected for 100ms prior to this point. Based on data from included participants only, the average number of accepted trials for rhyming letter pairs was 38.67 in children with CIs and 35.30 in children with typical hearing. The average number of accepted trials for the non-rhyming condition was 39.83 in children with CIs and 38.00 in children with typical hearing.

Six EEG channels, comprising Cz and the five channels directly adjacent to Cz, were included in analyses (see Figures 5.2 and 5.3). This distribution of channels was expected to capture the rhyme effect, based on previous research in which the rhyme effect is prominent in centro-parietal regions (Coch et al., 2011). As stated previously, electrical artefacts from CIs were not observed, though in order to further reduce the potential for contamination, it was considered important to restrict statistical analyses to predominantly midline electrodes, which were distal to the outer components of the CIs. This step is consistent with past research involving CI participants (e.g., Hahne, Wolf, Müller, Mürbe & Friederici, 2012). The specific region of interest, according to the 128-electrode GeoDesic sensor net system, included channels 7, 106, 31, 80, 55 and the reference channel. These channels respectively approximate C1, C2, Cp1, Cp2, Cpz and Cz in a 10-10 system. EEG data were averaged across these six electrodes for all analyses.

The mean amplitude of the rhyme effect for each participant was quantified by calculating the difference wave between the rhyming and non-rhyming conditions. Each participant's peak latency of the rhyme effect was measured as the latency of the most negative voltage value reached for the same difference wave. Visual inspection of the grand-averaged TH group's data revealed a large negative deflection between 300 and 500ms post-stimulus onset, peaking at a more negative amplitude for the non-rhyming condition; thus, this time region was considered suitable for rhyme effect analyses.

For the TH group, a paired samples t-test was used to compare mean amplitude values between rhyming and non-rhyming conditions. For Pearson's correlational analyses and case-control comparisons with CI participants, mean amplitude and peak latency values for the TH group's rhyme effect were obtained from the difference wave. For children with CIs, the limited power associated with the final cohort's sample size (n = 6) prohibited the use of group-based statistical comparisons. Accordingly, the waveforms generated for each participant in the CI group were examined visually, and with reference to results from case-control analytical methods. Data from the TH group served as a baseline with which to compare the mean amplitude and peak latency of the rhyme effect elicited in each CI group member. These ERP findings were also examined in the context of participants' behavioural results (on tests listed in Table 5.2), which were also subjected to the same case-control analyses. For each ERP value and behavioural test result, a *t*-value and associated two-tailed *p*-value were generated to indicate a statistically significant deviation from the TH group norms (alpha level = 0.05). The degree of deviation was quantified using effect size *z*-score values, which estimated the average difference, in standard deviation units, between a case's score and the score of a randomly selected member of the TH control group (Crawford, Garthwaite & Porter, 2010). All case-control analyses were conducted using the Singlims_ES computer program, designed by Crawford et al. (2010). All analyses for the CI group were computed based on the average of the six centro-parietal channels of interest (see Figures 5.2 and 5.3).

5.4. Results

5.4.1. TH group.

5.4.1.1. ERP results. The TH group's (n = 20) mean accuracy on the task was 88.95% (SD = 6.91%; rhyming = 85.08%; non-rhyming = 92.81%). To determine the presence of the rhyme effect for the TH group, a paired t-test was used to compare the mean amplitude of the rhyming and non-rhyming condition. The results confirmed that there was a significant rhyme effect, $t(19) = -2.206$, $p = 0.040$, such that the mean amplitude of the non-rhyming condition was more negative than the rhyming condition (see Figure 5.2).

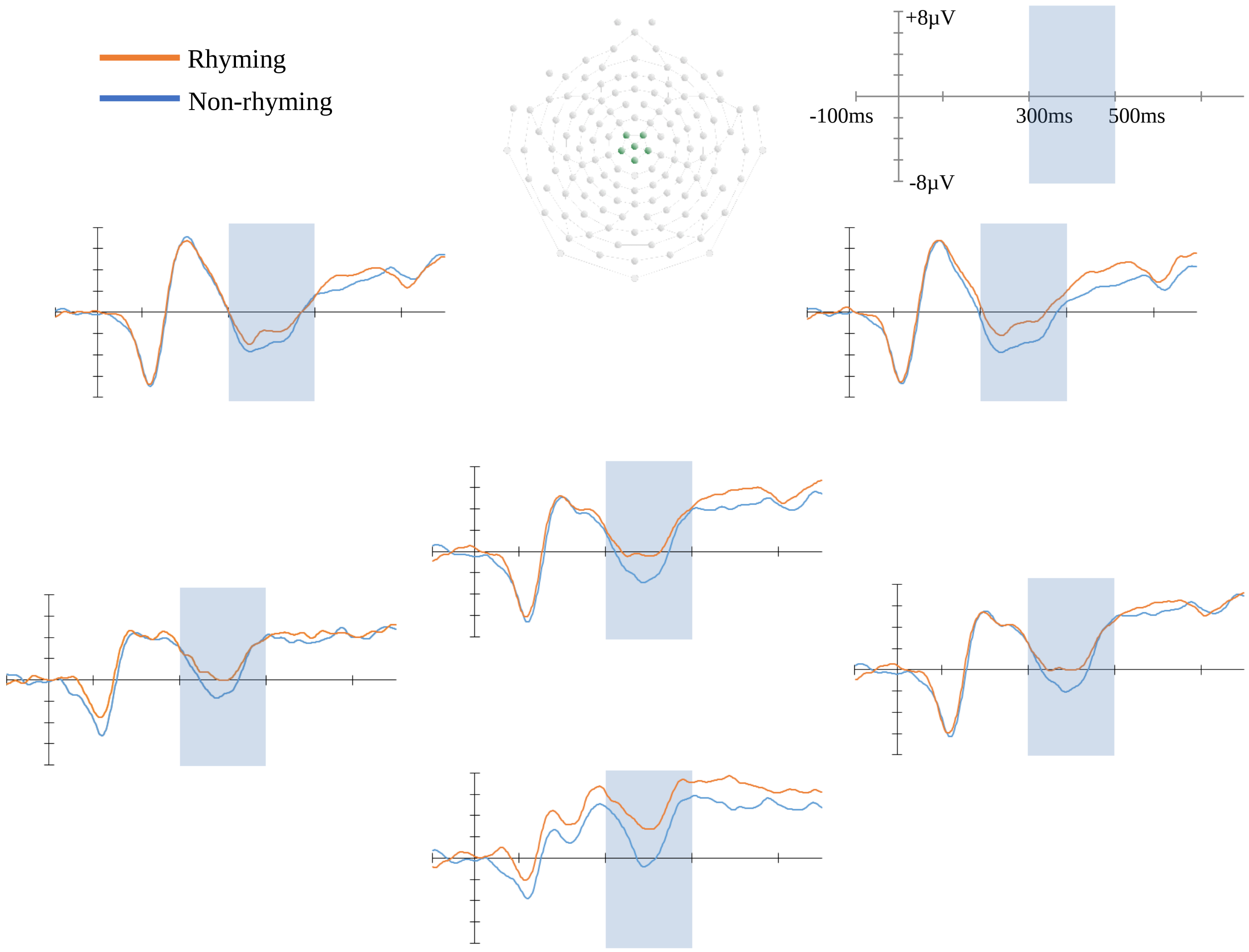

Figure 5.2. Grand-averaged ERPs for rhyming and non-rhyming conditions in typically hearing group.
Note. Shading used to indicate the time period after target onset (i.e., 300-500ms) captured in rhyme effect analyses. Six channels within centro-parietal region of interest were used in analyses (highlighted green in channel map).

5.4.1.2. Relationships between ERP and behavioural results. Detailed spoken and written language results for a larger participant cohort than that included here are reported elsewhere (see Chapters 2 and 3). A summary of the TH group's results pertaining to measures included in the present study's correlational analyses is provided in Table 5.2. Relationships between ERP and behavioural measures in the TH group were examined using Pearson's correlational analyses. The TH group's rhyme effect mean amplitude and peak latency values, calculated from the difference wave generated between rhyming and non-rhyming waveforms, were included in these analyses, as were behavioural measures of real word reading, nonword reading, letter-sound knowledge, phonological awareness, phonological memory, RAN and nonverbal reasoning.

A summary of the correlational results is given in Table 5.3. When unadjusted for nonverbal reasoning ability, accuracy on the letter rhyme judgement ERP task was correlated significantly with nonword reading, $r = 0.458$, $p = 0.042$, real word reading, $r = 0.458$, $p = 0.042$, and RAN, $r = 0.460$, $p = 0.041$, but not with the amplitude or latency of the ERP rhyme effect. Significant correlations were found between nonverbal reasoning and rhyme effect amplitude, $r = 0.534$, $p = 0.015$, and between letter-sound knowledge and rhyme effect amplitude, $r = 0.475$, $p = 0.034$. The correlation between phonological awareness and rhyme effect amplitude also approached significance, $r = 0.432$, $p = 0.057$. For all relationships between behavioural measures and rhyme effect mean amplitude, better performances were associated with smaller (i.e., more positive) rhyme effects. With respect to peak latency, the correlation between phonological memory and timing of the rhyme effect approached significance, $r = 0.438$, $p = 0.053$, such that a later latency of rhyme effect was associated with better phonological memory.

Although all participants were relatively homogeneous in their nonverbal reasoning abilities, given that they scored within or above the standardised age-based norms, the observed correlation between this measure and mean amplitude of the rhyme effect warranted further consideration. To examine whether nonverbal reasoning was driving the other correlational results, Pearson's correlations between behavioural measures and rhyme effect mean amplitude were re-computed with nonverbal reasoning ability partialled out (see 'adjusted' values in Table 5.3). As a result of implementing this step, the relationship between letter-sound knowledge and rhyme effect amplitude dropped to insignificance ($p = 0.323$).

Table 5.2

Typically hearing group's behavioural task performances (n=20).

Measure	Mean	Standard deviation
Real word reading[1]	0.87	1.42
Nonword reading[1]	0.77	1.29
Letter-sound knowledge[1]	0.67	0.87
Phonological memory[1]	0.36	0.92
Phonological awareness[2]	109.70	13.29
Rapid automatised naming[2]	102.25	9.98
Nonverbal reasoning[1]	1.30	0.88
Letter rhyme task accuracy[3]	88.95	6.91

Note. [1]Standardised z-score (normalised test mean = 0.0); [2]Standard Score (normalised test mean = 100.00). [3]Percentage correct across conditions.

Table 5.3

Correlations in typically hearing group (n=20).

		Real word reading	**Nonword reading**	**Letter-sound knowledge**	**Phonological memory**	**Phonological awareness**	**RAN**	**Nonverbal reasoning**	**LR accuracy**
RE amplitude	Unadjusted for nonverbal reasoning	.15	.07	*.48	.29	.43	.03	*.53	.25
	Adjusted for nonverbal reasoning	-.09	-.16	.24	.09	.26	-.01	-	.12
RE latency		.35	.22	.25	.44	.08	.38	.16	-.03
LR task accuracy		*.46	*.46	.23	.39	.38	*.46	.27	-

Note. RE = rhyme effect; LR = letter rhyme; RAN = rapid automatised naming. Statistically significant values (p<.05) indicated with asterisk (*).

5.4.2. CI group. The CI group's (n = 6) mean accuracy on the task was 82.81% (SD = 4.42%; median = 83.59%; range = 77.34-89.06%; rhyming = 78.13%; non-rhyming = 87.5%). Based on individual analyses, conducted separately for each participant from the CI group using standard case-control comparison procedures (Crawford et al., 2010), total task accuracy for children with CIs did not deviate significantly from that of the TH control group.

According to visual inspection of individually averaged waveforms, four participants from the CI group demonstrated a similar pattern of rhyme effect to that exhibited in the TH group (i.e., a more negative waveform generated for non-rhyming stimuli, compared with rhyming stimuli, within the 300-500ms time region). These participants (#1-4 in Figure 5.3) were examined first, followed by the other two participants who did not exhibit a similar pattern of rhyme effect to the TH group (#5 and #6 in Figure 5.3). Each participant's ERP and behavioural results were individually analysed, using the TH group's results as a comparative reference.

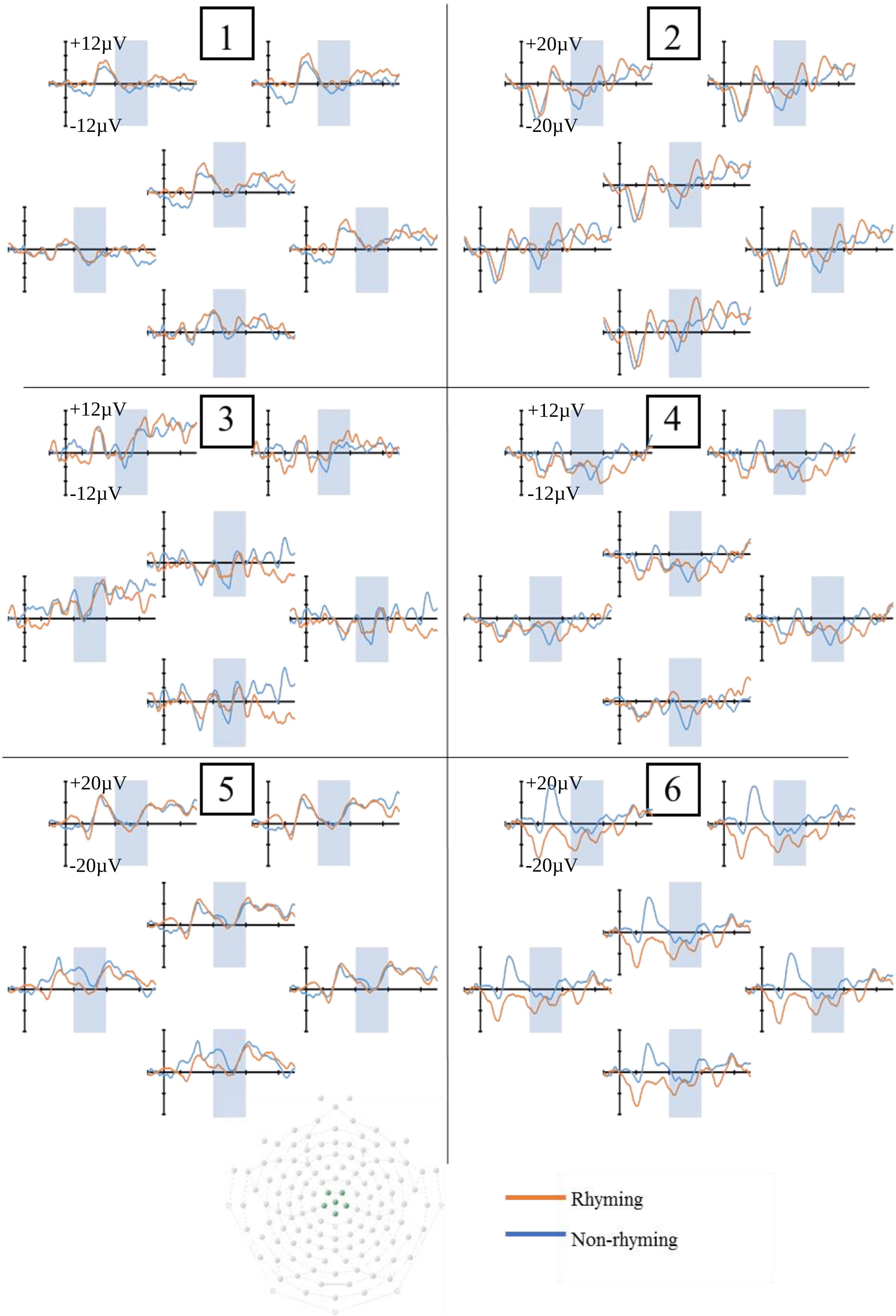

Figure 5.3. Rhyme effect in children with cochlear implants.
Note. Shading used to indicate the time period after target onset (i.e., 300-500ms) captured in rhyme effect analyses. Six channels within centro-parietal region of interest were used in analyses (highlighted green in channel map).

5.4.3. Participants with CIs: #1, 2, 3 and 4.

5.4.3.1. ERP results. Classical case-control difference tests revealed that the mean amplitudes and peak latencies of the rhyme effect in CI participants #1 through #4 were statistically similar to the TH group (all p-values > 0.3). In other words, the characteristics of the rhyme effect elicited in these individuals would not likely deviate from a randomly selected member of the TH control group. The results were supported by visual examination of the four participants. Although there appeared to be substantial between-subject variability in the amplitude of rhyme effect (e.g., #1 versus #2), the negative waveform peaks of rhyming and non-rhyming stimuli approximated the TH group's grand-averaged waveforms (Figure 5.2) in shape, latency and effect of condition.

5.4.3.2. Behavioural results. Participants #1 and #2 from the CI group performed similarly to the TH control group on all measures of reading and phonological processing. Nonverbal reasoning for participant #1 was above that of the TH control group, $z = 2.175$, $t = 2.123$, $p = 0.047$, while for participant #2, nonverbal reasoning did not deviate significantly from TH group norms. In the context of their ERP results, the successful elicitation of a rhyme effect may be related to a normalised behavioural assessment performance.

That said, while participant #3 also appeared to demonstrate a rhyme effect, as well as similar behavioural results to control group norms on most tests, their letter-sound knowledge was significantly below the TH mean, $z = -2.422$, $t = -2.363$, $p = 0.029$. Participant #4, who also demonstrated a rhyme effect, performed below the TH group on measures of phonological memory, $z = -2.868$, $t = -2.798$, $p = 0.011$, and RAN, $z = -2.629$, $t = -2.566$, $p = 0.019$. Nonverbal reasoning scores for participants #3 and #4 did not deviate significantly from the TH group norms.

5.4.4. Participants with CIs: #5 and 6.

5.4.4.1. ERP results. Based on visual inspection, CI participants #5 and 6 showed a reversed rhyme effect, whereby the waveforms elicited in response to rhyming stimuli were more negative than those elicited in response to non-rhyming stimuli. For participant #5, this appeared to be largely localised to the time window of interest (300-500ms), whereas for participant #6, the gap between conditions was broad and encompassed the entire time window. Statistical analyses confirmed that the mean amplitude of the rhyme effect elicited for participant #6 (6.00μV) deviated significantly from the TH control group (-1.29μV), $z = 2.785$, $t = 2.718$, $p = 0.014$. The peak latency of participant #6's rhyme effect (388ms) did not deviate significantly from TH norms (401ms). The mean amplitude of the rhyme effect for participant #5 (1.85μV) did not deviate from the TH norms to a statistically significant degree, and their peak latency of the rhyme effect (464ms) also did not deviate significantly from TH norms.

5.4.4.2. Behavioural results. On the behavioural assessment measure of nonverbal reasoning, CI participant #5's score was below that of the TH norms, but not significantly (p = 0.074). Participant #6's nonverbal reasoning score was significantly below TH norms, z = -2.336, t = -2.28, p = 0.034. Both children's letter-sound knowledge scores were significantly lower than the average of the TH control group norms (to the same degree), z = -2.203, t = -2.15, p = 0.045. No other phonological processing or reading scores deviated to a statistically significant degree.

5.4.5. Discontinuations and exclusions (both groups). Post hoc analyses were conducted to explore the high rate of participant discontinuations and exclusions in the present study. Data were collapsed across CI and TH groups, and independent samples t-tests were computed between participants who completed the task and achieved >70% accuracy (n = 27), versus those participants who did not complete the task, or who achieved <70% accuracy (n = 14). Variables of interest included age, nonverbal reasoning, nonword reading, real word reading, phonological awareness, phonological memory, RAN and letter-sound knowledge. Analysis of the results indicated that those who did not complete the letter rhyme judgement task (accurately) had significantly poorer nonverbal reasoning, $t(39)$ = 3.249, p = 0.002, nonword reading, $t(39)$ = 2.539, p = 0.015, real word reading, $t(39)$ = 2.488, p = 0.018, phonological awareness, $t(39)$ = 2.456, p = 0.019, phonological memory, $t(39)$ = 2.162, p = 0.037, and letter-sound knowledge, $t(39)$ = 3.375, p = 0.002, than those who did complete the task. There were no significant differences found in the ages or RAN scores of the participants who did and did not complete the task.

5.5. Discussion

The present study used a letter rhyme judgement paradigm to examine the rhyme effect in beginning readers with CIs. Due to task difficulty, results were obtained for only six children with CIs, and the reduced number of participants thus prohibited group-based analyses. Individual comparisons with the TH group were instead conducted, the results of which highlighted substantial variation in the presence and size of rhyme effect in children with CIs. The larger group of children with typical hearing produced a significant rhyme effect in response to the letter rhyme judgement task. Moreover, a smaller (i.e., more positive) rhyme effect amplitude in the TH group was correlated significantly with *better* letter-sound knowledge. Interpretations pertaining to observed results are described in detail below.

5.5.1. The rhyme effect in children with cochlear implants. The present study's approach to analyses allowed for a qualitative and quantitative examination of each CI participant's individual ERP waveforms, as contextualised by the results of the TH control group and those of other CI participants. Statistically, only one participant from the CI cohort deviated from the TH control group to a significant degree, in terms of the amplitude of their rhyme effect. These results suggest that, on the whole, the rhyme effect elicited in beginning readers with CIs did not diverge substantially from the normal distribution of rhyme effect elicited in TH children. Within the CI group, neural responses of participants varied considerably, although some patterns emerged that linked ERP results with behavioural assessment performances.

When children with CIs were sub-grouped according to whether or not they demonstrated a rhyme effect, the presence (or absence) of this effect appeared to be associated with nonverbal reasoning ability. Two children (#5-6 in Figure 5.3) showed a reverse rhyme effect – that is, the waveform elicited in response to rhyming letter pairs was more negative than the response to non-rhyming letter pairs. These same two children also demonstrated relative nonverbal reasoning difficulties. For participant #6, nonverbal reasoning was significantly below TH control group norms, while for participant #5, the deviance from TH norms only approached significance (p = 0.074), although both children were still within normal limits according to the standardised test norms. The finding that nonverbal reasoning was poorer for participants #5 and #6 (who did not show a visible rhyme effect) than for participants #1 through #4 (who did show a visible rhyme effect) suggests that nonverbal reasoning had some bearing on rhyme effect elicitation. However, further research is needed to confirm this assertion and establish the extent of this relationship, if any. It is possible that any link between rhyme effect amplitude and nonverbal reasoning will also be complicated by interactions with other cognitive-linguistic abilities, as appears to be the case with TH children (see Section 5.5.2).

Regardless of the above suggestion of a link between nonverbal reasoning and rhyme effect amplitude, the behavioural assessment performances did not appear to have an obvious or direct relationship with the visible characteristics of children's rhyme effects. For example, the two participants with CIs who performed best on behavioural assessment measures (i.e., #1-2 in Figure 5.3) produced distinctly different waveform and rhyme effect amplitudes. Such a contrast in neural responses elicited in otherwise similarly performing children serves to highlight the limitations of interpreting individual ERP data in isolation.

Given that, to the author's knowledge, this is the first study to investigate the rhyme effect in children with any degree of hearing loss, the preliminary findings presented here are unique. Yet, the claim that some children in the CI group demonstrated visible evidence of a rhyme effect is supported by results from MacSweeney and colleagues (2013), who examined adults with severe-to-profound hearing loss and also found a significant rhyme effect within this group. Results from the present study should therefore prompt further research into the rhyme effect in children with hearing loss, as examined on a group scale.

5.5.2. The rhyme effect in children with typical hearing. Children with typical hearing, who were aged between 6 and 9 years, demonstrated a significant rhyme effect in the present study, as evidenced by a more negative-going waveform in response to non-rhyming letter stimuli versus rhyming letter stimuli. The findings reported here are supported by previous studies in which letter stimuli were used to elicit the rhyme effect in adults (Bann & Herdman, 2016; Coch, George & Berger, 2008; Coch, Hart & Mitra, 2008; 2011; Stevens et al., 2013) and typically developing children (Coch et al., 2011). According to Coch and colleagues (2011), the two predominant steps involved in making a letter rhyme judgement are: (1) transcoding the letters (e.g., 'C') into phonological strings (e.g., /si/), and; (2) comparing the phonological strings of the prime and target letters (e.g., /si/ vs. /di/) to decide if they rhyme or not. The successful elicitation of a rhyme effect reported here (and by Coch et al., 2011) provides validation for using a letter rhyme paradigm with beginning readers, although correlational results from the present study also highlight potentially influential neural demands associated with the first 'transcoding' step.

Correlational analyses revealed a moderate and significant relationship between letter-sound knowledge and rhyme effect amplitude, such that better behavioural assessment scores were associated with smaller effects of condition. On the surface, the direction of this relationship appears counter-intuitive, since the rhyme effect is generally treated as an index of phonological sensitivity, and an individual's capacity for greater phonological sensitivity should be associated with *better* letter-sound knowledge (Wagner et al., 1994). Rather than being a spurious finding however, the observed correlation may instead represent the specific neural demands associated with the letter rhyme judgement paradigm. Before comparing a target letter's rime with that of its preceding letter stimulus, an examinee must retrieve the letter's phonological string – that is, its name. Theoretically then, competing information about a letter's (e.g., 'C') corresponding phoneme (e.g., /k/) may interfere with the process of letter name retrieval (e.g., /si/), thereby resulting in an attenuated effect of rhyme condition for those children with easier access to phonics knowledge. Other studies involving *word* rhyme judgement tasks have modulated the rhyme effect amplitude by manipulating the orthographic (Botezatu, Miller & Misra, 2015) and semantic (Perrin & Garcia-Larrea, 2003) characteristics of their stimuli. It may be that *letter* rhyme judgement responses are subject to similar occlusion effects, in this case resulting from the letter-sound characteristics of stimuli.

Importantly, the rhyme effect itself was still significant for TH children, which indicates its robustness to the potential influence of letter-sound knowledge. In addition, the letter rhyme paradigm has previously been used to elicit the rhyme effect in adults, who – although never explicitly tested – would presumably have better letter-sound knowledge than 6- to 9-year-old children. Hence, it is unlikely the observed correlation would extend to the point whereby those with excellent phonics skills show no rhyme effect whatsoever. Instead, the relationship between letter-sound knowledge and rhyme effect amplitude may be mediated by educational environment, since the present study's cohort of beginning readers were likely receiving phonics-based literacy instruction, and this may have heightened the ease with which letter-sound knowledge was retrieved. Interestingly too, the correlation between letter-sound knowledge and rhyme effect amplitude disappeared when nonverbal reasoning was statistically controlled. This finding suggests that children with good nonverbal reasoning skills may be better equipped to cope with the neural demands of competing letter-name versus letter-sound information. Further research may elucidate the extent to which modulatory factors, such as learning environment and nonverbal reasoning ability, influence the relationship between letter-sound knowledge and the letter rhyme effect.

According to correlational results from the present study, accuracy on the letter rhyme judgement task was significantly correlated with real and nonsense word reading ability, as well as RAN. In support of these findings, Coch et al. (2011) found that letter rhyme judgement accuracy was correlated with real/nonsense reading accuracy, letter name knowledge and phonological awareness percentile rank scores (but not standard scores) in 6- to 9-year-old children. Together, the results strongly suggest that at a behavioural level, participants' responses to the letter rhyme judgement task rely on skills that also underlie reading development, such as those involved in converting orthographic information to a corresponding phonological string.

5.5.3. Letter rhyme judgement: limitations and future directions. There is evidence from the present study that participants struggled with the cognitive-linguistic demands associated with letter rhyme judgement. Firstly, a substantial proportion (n = 14) of the original participant cohort (n = 41) did not complete the task or were excluded because of inaccuracy. This outcome is unlikely to be an artefact of non-compliance with EEG testing, since most of the cohort successfully completed one other ERP task on the same day as the letter rhyme judgement task. Participant fatigue is also an unlikely factor, because the order of the two ERP tasks was balanced within each TH and CI group. Similarly, the number of participant exclusions due to non-completion was roughly equivalent across those who completed the rhyme judgement task first, versus those who completed it second. As described earlier, the two main steps involved in completing the letter rhyme judgement task (according to Coch et al., 2011) are: (1) transcoding the letter symbol into its name, and; (2) comparing the names (or phonological strings) of the two letters, to determine rhyming status. When tested behaviourally, all participants demonstrated high letter name accuracy (albeit on an untimed and informal measure), and so the high rate of participant exclusions cannot be attributed to an inability to perform step 1. It is possible, since rhyme awareness was not explicitly assessed during testing, that the children did have difficulty performing step 2. This assumption is supported by the finding that children who could not complete the task accurately performed significantly worse on measures with high phonological processing demands (e.g., phonological awareness, phonological memory, and reading).

Moreover, any relative phonological processing weaknesses may have been exacerbated by the conceptual challenge of judging rhyme in *letters*. Again, this assumption is supported by group difference analyses, the results of which revealed significantly poorer nonverbal reasoning abilities in children who did not complete the letter rhyme task. As mentioned previously, the age and curricular expectations of beginning readers in the present study were such that all children were probably receiving literacy instruction at school with an emphasis on phonics skill development. For those with comparatively low cognitive processing abilities, it may therefore have been more difficult to cope with the unfamiliar neural demands associated with letter rhyme judgement, wherein the task is to retrieve and process the phonological structure of a letter's name, rather than the letter's corresponding sound.

In summary, a limitation of the present study was that some participants with nonverbal and linguistic processing difficulties could not be included in the actual ERP analyses, because of the nature of the ERP task itself. Given that neither discontinuations nor exclusions on the basis of inaccuracy were reported by Coch et al. (2011), the present study's high exclusion rate may be associated with an Australian (versus United States) educational system, and the literacy-based curricular targets therein. Further research is warranted to explore the relationships between letter-sound processing, nonverbal reasoning and both ERP and behavioural characteristics of the letter rhyme effect. To this end, it may be interesting to examine the executive functioning and attentional demands associated with eliciting the rhyme effect in children, in order to better understand the extent to which cognitive factors influence ERP results.

5.6. Conclusion

In the present study, beginning readers with CIs and beginning readers with typical hearing performed a letter rhyme judgement task while their ERP activity was recorded. A significant rhyme effect was elicited in the TH group, and – based on visual inspection of waveform data – in some individuals with CIs. For TH children, a smaller amplitude of rhyme effect was significantly correlated with better letter-sound knowledge, although this relationship disappeared when nonverbal reasoning abilities were statistically accounted for. Hence, potential interference effects associated with the retrieval of stimulus *phonemes* (as opposed to *names*) may have been mediated by higher nonverbal functioning. The letter rhyme judgement task appeared to involve high cognitive-linguistic processing demands, as evidenced by the high participant exclusion rate: this may be a fruitful area of investigation for research into the methodological constraints of rhyme effect elicitation. Future studies that include children with CIs may also examine the rhyme effect on a group scale, so that inferences can be made about the neural functioning of their underlying phonological skills.

Chapter 6.

General Discussion

6.1. Introduction

The research presented in this thesis has provided valuable insight into how literacy skills develop in children with cochlear implants (CIs). Already, there is a substantial body of existing literature to suggest that individuals with severe-to-profound hearing loss demonstrate spoken and written language processing difficulties, when compared with their typically hearing peers (see Harris, 2015, for review). Yet, the degree to which such findings can be generalised to specific subsets of the hearing-impaired population is limited by the heterogeneity of participants sampled, with respect to their age at testing, form of aiding, exposure to intervention, and communication mode. The aim of this thesis was to examine in depth the literacy development of beginning readers with CIs, who use only spoken communication and who received early auditory-verbal therapy (AVT) behavioural intervention.

In Chapter 1, the existing literature pertaining to literacy development in children with CIs was reviewed, in order to provide context for the research conducted in Chapters 2 to 5. In Chapters 2 and 3, behavioural assessment measures were used to examine, respectively, reading and spelling in children with CIs. In Chapters 4 and 5, electroencephalography (EEG) measures were used to examine the neural functioning of underlying literacy-related skills. Semantic processing was the focus of Chapter 4, while phonological processing was the focus of Chapter 5. A common thread linking all chapters in the thesis thus far has been the psycholinguistic nature of the research, with respect to methodological design, and the analysis and interpretation of data. Hence, particular attention will be devoted in Chapter 6 to applying components of psycholinguistic-centred theoretical models of word- and text-level written language processing to the reported results.

Following a summary of the key thesis findings, skills underlying literacy development will be discussed in detail, with reference to how children with CIs performed on measures of phonological, orthographic and semantic processing, and to what degree these abilities contributed to the children’s word-level reading and spelling. The discussion will then move from word-level to text-level literacy development and the contribution of underlying skills to overall reading comprehension. Finally, limitations of this research project will be described, as will possible future directions for further investigation.

6.2. Key Findings

The following is a summary of the key findings obtained from Chapters 2 to 5 of the present thesis:

1. Children with CIs performed significantly worse than typically hearing (TH) age-matched controls on behavioural assessment measures of word- and text-level reading accuracy, although their text-level reading comprehension did not differ significantly to controls.
2. For both CI and TH groups, listening comprehension and word-level reading accuracy contributed significantly to text-level reading comprehension. The predominant concurrent predictor for the CI group was word-level reading accuracy, while the predominant concurrent predictor for the TH group was listening comprehension.
3. For both CI and TH groups, phonological and orthographic processing skills contributed to word-level reading accuracy, with orthographic processing as the predominant concurrent predictor for both groups.
4. Children with CIs and TH children exhibited similar performances on behavioural assessment measures of irregular and nonsense word spelling accuracy.
5. Children with CIs produced significantly fewer phonologically plausible spelling errors than children with typical hearing.
6. For both the CI and TH groups, orthographic processing skills contributed to irregular word spelling accuracy.
7. Letter-sound knowledge did not contribute significantly to nonword spelling accuracy for the CI group, whereas it did for the TH group.
8. Children with CIs demonstrated normal sensitivity to lexical-semantic incongruence, as indexed by an N400 effect which was similar in size and shape to that produced by TH children.
9. Some children with CIs demonstrated a rhyme effect similar to TH children, based on visual inspection and individually conducted analyses.
10. For both the CI and the TH children, the rhyme effect elicited in response to a letter rhyme judgement task may have been influenced by participants' letter-sound knowledge and nonverbal reasoning ability.

6.3. Skills Underlying Word-level Literacy Development in Children with Cochlear Implants

Existing models of skilled word reading account for the contributions of underlying phonological (i.e., speech sound-based), orthographic (i.e., letter-based), and semantic (i.e., meaning-based) processes. According to the dual-route cascaded (DRC) model, words are decoded or recognised via different reading 'routes' (Coltheart, Rastle, Perry, Langdon & Ziegler, 2001). Unfamiliar words are processed via a sublexical route, in which the reader applies existing knowledge of letter-sound correspondences to 'sound out' items, thus relying on a functional phonological system (Coltheart et al., 2001). Conversely, familiar words are recognised as whole orthographic representations via a lexical route, which essentially bypasses the phonological system and allows for understanding of word meaning through access to the adjacent semantic system (Coltheart et al., 2001).

An alternative, connectionist perspective on single word reading is theorised in the parallel distributed processing (PDP) model, which assumes that orthographic, semantic and phonological representations for a given item are simultaneously computed (Seidenberg & McClelland, 1989). Rather than taking different routes, all types of letter strings are recognised with input from the same underlying components. Yet, the degree to which each component of the model contributes to the outcome is separable, as with the DRC model.

Although the DRC and PDP theoretical models differ with respect to the exact processes by which successful word recognition is thought to be achieved, they agree that the same three underlying areas (i.e., phonology, orthography and semantics) are fundamentally involved. In the present thesis, phonological, orthographic and semantic processing abilities were assessed in children with CIs and in children with typical hearing. The specific purpose of examining these areas was to determine how such processes – both together and in isolation – contributed to word-level literacy development.

6.3.1. Phonological processing.

6.3.1.1. Phonological processing skills in children with cochlear implants. Throughout this thesis, the term 'phonological processing' was used to refer to the explicit or implicit computation of speech sound representations, in the form of storing, accessing, retrieving or manipulating such information, or otherwise making use of it to decode and encode written language (Wagner, Torgesen & Rashotte, 1994). Prior studies involving children with CIs have consistently reported poorer outcomes for this cohort when they perform tasks that have high phonological processing demands (Ambrose, Fey & Eisenberg, 2012; James, Rajput, Brinton & Goswami, 2008; Lee, Yim & Sim, 2012; Nittrouer, Caldwell, Lowenstein, Tarr & Holloman, 2012; Nittrouer, Caldwell-Tarr, Sansom, Twersky & Lowenstein, 2014; Spencer & Tomblin, 2009; Weisi et al., 2013). Indeed, such evidence aligns with the findings outlined in Chapter 2 of this thesis, in which beginning readers with CIs demonstrated significantly poorer phonological awareness, rapid automatised naming (RAN), letter-sound knowledge and phonological memory than TH children. In particular, children with CIs showed substantial difficulty performing the nonword repetition task, which was used to measure phonological memory.

As described in Chapter 5 of this thesis, phonological processing was examined on a neurophysiological level, as well as on a behavioural level. Past studies have used EEG to examine rhyme sensitivity as an index of broader phonological processing. Specifically, an event-related potential (ERP) elicited 300 to 600 milliseconds after a target stimulus is found to be more negative if the target is preceded by a non-rhyming item, relative to a rhyming item (Grossi, Coch, Coffey-Corina, Holcomb & Neville, 2001). This effect of rhyming versus non-rhyming conditions is referred to as the ‘rhyme effect’. As yet, there is no evidence pertaining to the rhyme effect in children with CIs. The results from Chapter 5 indicated that, on an individual level, some children with CIs produce a visible rhyme effect in response to letter stimuli. There was also substantial variability among CI participants with respect to the size and shape of ERP waveforms produced, although neither the amplitude nor the latency of the rhyme effect had an obvious correspondence with these individuals’ behavioural responses to phonological processing tasks. Interestingly, a link between rhyme effect amplitude and behavioural letter-sound knowledge did emerge for the TH control group, and this warrants future investigation, as detailed in Section 6.6.

6.3.1.2. Phonological processing as a literacy sub-skill in children with cochlear implants. Since the DRC and PDP models described in Section 6.3 represent *skilled* word recognition, age-related changes to the predictive value of each underlying system are not explicitly considered. That said, repeated and early exposure to letter-sound associations (e.g., through phonics training) is found to be conducive to the establishment of increasingly refined orthographic representations (Harm & Seidenberg, 2004). The role of phonology is therefore expected to be comparatively increased in beginning readers, whose orthographic representations are still developing (Harm & Seidenberg, 2004). This hypothesis is supported by findings from longitudinal studies with young TH children, in which early phonological awareness skills are predictive of later reading outcomes (Badian, 1998; Hulme et al., 2002; Wagner et al., 1994). In addition, both children and adults with dyslexia are commonly observed to perform poorly on phonological tasks, and the ‘phonological core deficit’ hypothesis for dyslexia attributes word-level literacy difficulties to a dysfunctional phonological system (Stanovich, 1988).

Previous studies involving children with CIs have found a longitudinal relationship between early pre-literate phonological awareness performance and school-age word reading performance (Harris & Beech, 1998; Colin, Magnan, Ecalle & Leybaert, 2007). Such findings suggest that phonological skills are important for reading development in hearing and non-hearing children alike (Mayer & Trezek, 2014). However, there remains considerable debate on this point. A meta-analysis by Mayberry, del Giudice and Lieberman (2011) reported that only 11% of the variance in reading ability for individuals with varying degrees of hearing loss was explained by concurrent phonological measures, while 35% was explained by language ability. The authors thus concluded that phonological skills are not essential in attaining successful reading outcomes, and word learning may instead be achieved on the basis of linguistic knowledge and holistic orthographic information. Notably, the participants included in Mayberry et al.’s (2011) meta-analysis ranged from 4 to 62 years of age, the breadth of which does not account for the changing contributions of causal reading skills over time. In addition, the actual proficiency of reading skill for the group with hearing loss was not reported, which makes it unclear whether age-appropriate reading achievement can be attained in the absence of adequate underlying phonological ability.

On the basis that children with CIs in the present study were all beginning readers (i.e., in the first three years of formal literacy instruction), it was hypothesised that phonological awareness would contribute significantly to their word reading outcomes. This question was addressed in Chapter 2 using multiple regression analyses, the results of which indicated that phonological awareness did indeed contribute variance to word reading accuracy for the CI (β = 41.0%, p = 0.006) and TH groups (β = 34.1%, p = 0.010). The disparity between these results and the 11% reported by Mayberry et al. (2011) is likely to have resulted from the much younger (and less variable) age of our cohort, and our specific focus on the relationship between phonological awareness and word-level reading accuracy, rather than the relationship between general phonological and reading measures. Given the cross-sectional nature of the study, it cannot be concluded from Chapter 2’s results that phonological awareness was a critical precursor skill that facilitated literacy growth in these children. Indeed, it is likely that, as has been found in other studies of typically developing children (e.g., Wagner et al., 1994), the causal relationship between phonological awareness and reading was bidirectional. Nevertheless, the findings provide support for a significant relationship existing between phonology and early reading development in children with CIs.

Chapter 3 examined the relationships between phonology and spelling achievement. Past studies have found that children with CIs produce fewer phonologically plausible errors (i.e., homophones or pseudohomophones of the target word) than TH children (Harris & Terlektsi, 2011; Hayes, Kessler & Treiman, 2011; Roy, Shergold, Kyle & Herman, 2015). Children with CIs in the present thesis conformed to the same pattern of results, producing 36.11% phonologically plausible errors in contrast to the TH group's 48.59% phonologically plausible errors. Interestingly, the CI group's total misspellings consisted of only 13.19% 'phoneme omission' errors, which was roughly equivalent to the TH group (12.53%). Hence, all children appeared to be largely aware of the phonemic structure of target words, although the children with CIs may have had difficulty representing that awareness with phonologically (and morphologically) legal letters. As was also described in Chapter 3, regression analyses were computed to determine the contribution of underlying phonological skills to nonword spelling accuracy. For TH children, letter-sound knowledge contributed significant variance to nonword spelling performance, whereas this relationship was non-significant for the CI group. Taken together, results from the misspelling analysis and regression profiles suggest that phonological – and more specifically, phonics – skills contribute less to spelling outcomes for children with CIs, when compared with TH children.

On the surface, the results discussed so far appear to indicate that phonological processing abilities contribute significantly to reading outcomes in children with CIs, but not to their spelling outcomes. This discrepancy is likely related to differences in the specific phonological processing skills measured in each study. With respect to reading, the phonological measure of interest was phonological *awareness*, while for spelling, it was the absence of a relationship between *phonics* knowledge and spelling outcomes that was most striking in the CI group. In the context of both studies, the results suggest that children with CIs can draw on phonological awareness skills (i.e., skills in manipulating spoken word parts) when decoding words, but cannot readily apply phonics knowledge (i.e., knowledge of how phonemes are represented by letters) to encode single words and nonwords.

6.3.2 Orthographic processing.

6.3.2.1. Orthographic processing skills in children with cochlear implants. In addition to phonological processing, the research presented in this thesis included an examination of orthographic processing. 'Orthographic processing' refers to the acquisition, storage and use of specific written word representations, and the understanding of the patterns and conventions governing our written language system (Apel, 2011). A beginning reader's orthographic processing skills are presumed to develop at least partly via repeated exposure to the correspondences between letter sequences and phonemic representations (Share, 1995). However, once learned, a word may be recognised on the basis of its orthographic representation, rather than on the basis of its letter-phoneme constituents. Orthographic processing is thus distinct from phonological processing.

Children with CIs in the present study performed well on the behavioural measure of orthographic processing. As reported in Chapters 2 and 3, the CI group's average orthographic processing z-score did not differ significantly from the TH group. Hence, despite the observed phonological processing difficulties, children with CIs could adequately access the orthographic representations of words. This finding suggests that orthographic skills can develop at least somewhat independently from phonological processing, and such skills can thus contribute unique variance to reading and spelling outcomes.

6.3.2.2. Orthographic processing as a literacy sub-skill in children with cochlear implants. For both children with CIs and children with typical hearing, orthographic processing contributed significantly to word reading outcomes (see Chapter 2). While not directly compared, the value of this contribution did not appear to differ substantially between groups, suggesting that the CI group did not 'compensate' for phonological deficits by relying more so than TH children on the retrieval of whole-word orthographic information. That said, overall word reading accuracy was poorer for CI children, which suggests their reading development was delayed to some degree in comparison to the chronological age-matched control group. It would be interesting for future research to conduct a similar examination using a *reading* age-matched control group, to determine whether the same word reading outcomes are achieved on the basis of different underlying processes, depending on group status. It is possible that the children with CIs relied more so on orthographic processing than would a younger TH cohort at a similar stage of literacy development.

Multiple regression analyses with spelling performance as the dependent variable were conducted with irregular words and nonwords examined separately (see Chapter 3).

Irregular words, by definition, contain uncommon letter-sound correspondences, and must be recognised and spelled using knowledge of existing orthographic information (Houghton & Zorzi, 2003). It is perhaps unsurprising that orthographic processing contributed significantly to irregular word spelling outcomes for children in both the CI and the TH groups. Given that the study presented in Chapter 3 is the first to examine such a relationship, the similarity in profiles across groups is interesting to observe, especially since irregular word spelling accuracy was equivalent between groups.

To summarise, the results from this thesis indicate that children with CIs were as competent as TH children when processing orthographic information, and such skills contributed to a similar degree to word-level reading and spelling outcomes. Possibly, the CI group relied on orthographic processing more strongly than would a younger TH cohort at a similar stage of reading development, and this supposition warrants further research. The strong relationship between orthographic processing and word-level literacy development may have resulted in the CI group's comparative success processing irregular words (see Section 6.4), although, overall, word- and text-level reading accuracy scores were still poorer for children with CIs than for TH children.

6.3.3 Semantic processing.

6.3.3.1. Semantic processing in children with cochlear implants. 'Semantic processing' refers to the computation of word meaning. In Chapters 2 and 4, children's receptive vocabulary skills were assessed and compared between CI and TH groups. To complete this task, participants were required to access the semantic properties of a spoken word in order to match it with one of four given pictures. Children with CIs performed significantly poorer than the TH control group, although their skills in this area were, on average, comparable to the standardised test sample (mean = 99.83; SD = 17.55). Participants were also assessed on their ability to understand and carry out spoken sentence-level directions, and on their comprehension of spoken passage-level texts. Both sentence- and passage-level tasks necessitated semantic processing at a single word level, but also required the integration of this knowledge with surrounding contextual information. In the sentence-level comprehension task, children with CIs performed significantly poorer than the TH control group, although the groups' scores were similar on the passage-level comprehension task (see Chapter 4).

Semantic processing was examined in further depth in Chapter 4, wherein participants completed a word-picture matching task to elicit an ERP component called the ‘N400’. For both CI and TH groups, the N400 elicited in response to non-matching word-picture pairs was more negative than that in response to matching word-picture pairs; hence, the groups showed a significant ‘N400 effect’. The amplitude and latency of this effect was similar across groups, indicating that the lexical-semantic processing skills indexed by the N400 effect were not especially deviant in the CI cohort. The study reported in Chapter 4 is unique, with respect to its focus on the specific subset of hearing-impaired children, who have CIs and use spoken language. The findings are, however, supported by previous research findings in which a significant N400 effect was demonstrated in younger children with CIs, who used a mix of sign and spoken language (Kallioinen et al., 2016; Vavatzanidis, Murbe, Friederici & Hahne, 2018).

Given that children with CIs demonstrated poorer vocabulary and spoken language abilities than TH children, there was evidently some dissociation between the semantic processing skills measured by behavioural versus ERP measures in Chapter 4. Possibly, the behavioural spoken language performance discrepancies between participant groups were not substantial enough to be reflected on a neural level, at least with respect to elicitation of the N400 effect. Indeed, past studies that have found reduced or delayed N400 effects in certain clinical populations have focused on those with diagnosed language disorders (Cummings & Ceponiene, 2010; Pijnacker et al., 2017). Children with CIs, in contrast, performed comparably with behavioural spoken language standardised test norms.

6.3.3.2. Semantic processing as a literacy sub-skill in children with cochlear implants. As with phonological and orthographic processing, the present study sought to determine the degree to which computation of word meaning contributed to word-level reading outcomes in children with CIs. According to the results reported in Chapter 2, semantic processing ability, as indexed by behavioural receptive vocabulary performance, was significantly related to word reading accuracy in CI and TH groups (see Chapter 2). An important caveat to these results was that vocabulary failed to contribute significantly to word reading accuracy when included in the same regression model as phonological and orthographic processing measures. Hence, although related to reading development in both groups, vocabulary was also highly correlated with other underlying processing abilities.

With respect to Chapter 4’s ERP measurement of semantic processing, no significant correlations were found between the N400 effect and behavioural word reading accuracy.

Given the aforementioned behavioural vocabulary-reading relationship, the absence of significant correlational results in Chapter 4 does not indicate that semantic processing is an unimportant factor in facilitating either participant group's reading development. Rather, behavioural performance on a word reading measure represents the end-point of numerous underlying skills, and the very fine-grained measurement of semantic sensitivity, recorded within half a second of exposure to a matching or mismatching target stimulus, was likely too indirectly related to children's word reading outcomes to appear significant, at least within the relatively small participant groups examined in this thesis. Although other studies have reported significantly deviant N400 effects in populations with diagnosed reading deficits (Jednoróg, Marchewka, Tacikowski & Grabowska, 2010; Stelmack & Miles, 1990), average word reading accuracy for Chapter 4's CI group was within normal limits (mean z-score = -.037; SD = 1.00), and this may have accounted for the finding that N400 effects were similar across CI and TH groups. In the context of previous research therefore, the results suggest that greater variability in reading performance (across and within CI and TH groups) may be associated with a greater chance of finding a significant relationship between ERP and behavioural reading outcomes.

6.4. Word-level Literacy Development in Children with Cochlear Implants

The word reading and spelling performances of CI and TH groups were examined in Chapters 2 and 3, respectively. Overall word-level reading accuracy, as indexed by a composite of regular, irregular and nonword reading accuracy scores, was significantly poorer for children with CIs than children with typical hearing. This finding was supported by results from previous research, in which school-age children with CIs who used spoken communication showed word-level reading deficits (e.g., Nittrouer et al., 2014; Weisi et al., 2013). Unlike these studies however, the present CI cohort was homogeneous in terms of early exposure to AVT and having normal nonverbal reasoning.

Spelling accuracy was examined in terms of participants' written productions of irregular and nonsense word stimuli. For both word types, children with CIs performed similarly to TH children. Such a similarity in spelling scores between CI and TH groups does not align with previous studies that have reported significant difficulties in children with CIs (Apel & Masterson, 2010; Geers & Hayes, 2010; Roy et al., 2015). That said, better outcomes have been reported in studies involving children with CIs who also use only spoken communication (e.g., Fitzpatrick et al., 2012; Hayes et al., 2011). Thus, the relatively positive spelling accuracy outcomes for children with CIs may be associated with their use of spoken communication and the early intervention which promoted such an approach to communication.

An integral aspect of the present thesis was that different types of word stimuli were used to measure participants' reading (Chapter 2) and spelling (Chapter 3) accuracy. According to existing theories of single word processing, as described in Section 6.3 of this chapter, different underlying deficits may affect performance differentially across word types[1]. In this regard, it is worth noting that for both reading and spelling, children with CIs appeared to perform better when processing irregular words, compared with nonwords. The CI group performed significantly worse than the TH group on nonword reading accuracy (CI mean = -.32; SD = .91; $p = .025$) and regular word reading accuracy (CI mean = -.28; SD = 1.13; $p = .036$), while the group difference for irregular word reading accuracy only approached statistical significance (CI mean = -.14; SD = 1.29; $p = .054$). Similarly, nonword spelling accuracy for the CI group was poorer than the TH group to a degree that approached significance (CI mean = -.05; SD = 1.25; $p = .064$), while irregular word spelling accuracy was equivalent across groups (CI mean = .04; SD = 1.25; $p = .467$). Importantly, reading and spelling accuracy as a factor of word type was not investigated explicitly, which means that although the appearance of better irregular than nonsense word reading and spelling accuracy is indicative of the CI group's impaired phonological processing relative to their orthographic processing, the findings are not conclusive. Further research in this area is warranted, and in particular, it would be useful for future studies to compare the developmental outcomes of children with CIs exposed to phonologically- versus orthographically-focused interventions.

Even if word type did factor into the CI group's word-level literacy performances, it is worth noting the different degrees of success observed in children with CIs, with respect to their reading and spelling. That is, the CI group demonstrated poorer single word reading skills than TH controls, but similar single word spelling skills. Such a pattern of results has also been found in past studies involving children with severe or profound hearing loss (e.g., Aaron, Keetay, Boyd, Palmatier & Wacks, 1998; Burden & Campbell, 1994; Colin, Leybaert, Ecalle & Magnan, 2013; Harris & Moreno, 2004). According to Roy and colleagues (2015), applying non-phonological skills to the processing of written words does not affect spelling

[1] The PDP model is based on statistical learning principles. As such, it treats all lexical items as points on a continuum of spelling-sound consistency (although proponents of the model do still make a distinction between 'rule-governed' and 'exception' words; Seidenberg, 2005). The question of partial regularity is beyond the scope of this thesis; instead, regular (i.e., rule-governed) and irregular (i.e., exception) words may be considered exemplars of the extreme ends of the spelling-sound consistency continuum.

as much as reading, since spelling relies more on the retrieval of existing orthographic representations than on the application of sublexical phonological skills. Hence, even if reading and spelling development for children with significant hearing loss is based to a similar degree on non-phonological skills, such a route may be less problematic for spelling performance.

Alternatively, the observed divergence between the CI group's literacy outcomes may be attributed to their drawing on separate skills to read and spell words. Kyle and Harris (2011) reported that the vocabulary and speechreading skills of 5 year olds with severe or profound hearing loss significantly predicted reading performance two years later. In contrast, letter name knowledge was the only significant predictor of their spelling. A different pattern of results emerged for TH children matched on reading ability: speechreading and phonological awareness at 5 years predicted later skills in both reading and spelling, suggesting these outcomes were intrinsically linked. Interestingly, Kyle and Harris (2011) also found a strong correlation between the spelling and reading accuracy of their 5-year-old cohort with hearing loss. This and other similar findings (e.g., Harris & Terlektsi, 2011; Roy et al., 2015) indicate the developmental trajectories of reading and spelling are based on at least some of the same underlying skills.

6.5. Text-level Reading Development in Children with Cochlear Implants

Beyond just the single word level of written language processing, results from this thesis (described in depth in Chapter 2) also pertain to text-level reading abilities. The children with CIs were significantly less accurate than TH controls in terms of reading aloud passages of written text, although there were no significant group differences in text reading rate or comprehension. The similarity in reading comprehension scores between CI and TH groups was unexpected, since much of the existing literature points to difficulties in this area for children with CIs (Geers, 2003; Nittrouer et al., 2014; Spencer, Barker & Tomblin, 2003; Weisi et al., 2013). That said, Wu et al. (2013) also found similar reading comprehension abilities across CI and TH children. The comparatively positive reading comprehension results reported in Chapter 2 may be attributable to the CI group's high nonverbal IQ (mean z-score = .72; SD = 1.21) and the early age at which they received their CIs (mean z-score = 1.74y; SD = 1.45y).

Based on the Simple View of Reading model, reading comprehension is the product of word recognition and listening comprehension (Catts, Hogan & Adlof, 2005; Catts et al., 2015; Hoover & Gough, 1990). Indeed, both of these skill areas contributed significantly to reading comprehension for the CI and TH groups studied in Chapter 2. The groups' profiles differed somewhat from one another, with respect to the predominant concurrent predictor within the regression models. For the children with CIs, word recognition (i.e., word reading accuracy) contributed most to reading comprehension, while for children with typical hearing, listening comprehension (i.e., comprehension of spoken paragraphs) contributed most.

The difference in reading comprehension profiles between groups may have reflected the TH children's comparatively higher degree of word recognition automaticity, and their ability therefore to devote more cognitive resources to comprehending text meaning. Indeed, studies with typically developing children have found that spoken language ability contributes increasingly to reading comprehension with age, as decoding skills advance and word-level recognition becomes more efficient (Language & Reading Consortium, 2015). Thus, in the context of their other results, the phonological processing deficits demonstrated by children with CIs appear to have flow-on effects to word-level reading development, and these difficulties appear also to influence the children's text-level reading profiles, though not to the extent that reading comprehension is in turn, as yet, impaired. Importantly, the analyses reported in this thesis focused only on beginning readers in the first few years of formal schooling, so it may be the case that reading comprehension difficulties present later on, when the academic demands are such that these children are expected to 'read to learn', rather than 'learn to read'. Accordingly, the trend in other studies is for the performance gap between children with hearing loss and children with typical hearing to widen with age (Geers & Hayes, 2010; Geers, Tobey, Moog & Brenner, 2008).

6.6. Limitations and Future Directions

Children with hearing loss in the present study were relatively homogeneous, in terms of their age at assessment, the severity and laterality of their hearing loss, their use of CIs and spoken language, and the normality of their nonverbal reasoning abilities. As such, the results reported in the present thesis cannot necessarily be generalised to children with hearing loss who do not meet the same criteria. Further research may benefit from the inclusion of a more heterogeneous CI sample, assuming of course that the size of CI group in such research is sufficient for multiple factors to be statistically controlled. For the research presented in this thesis, group-based comparisons involving children with CIs were conducted based on a sample size of 12 (Chapter 4) or 14 (Chapters 2 and 3). Thus, future studies should aim to increase the statistical power of their multi-factorial group-based analyses by examining a larger CI cohort.

Sample size was a particular concern for the letter rhyme EEG task (Chapter 5), wherein only six participants with CIs completed the task with adequate accuracy. As a result, statistical analyses were limited to individual case-control comparisons, rather than CI versus TH group comparisons. There is then certainly the impetus for future research to investigate the rhyme effect in children with CIs (and hearing loss more broadly), with a large enough sample size to allow for more powerful group-based analyses. A related limitation of the letter rhyme study described in Chapter 5 was that some children from CI and TH groups were unable to complete the task. This finding may be attributed to the conceptual and/or linguistic challenges associated with the task itself, since nonverbal reasoning, reading and phonological processing skills were significantly poorer in the subset of participants who did not complete the task or who achieved less than 70% accuracy. Hence, all analyses were conducted with only those participants from CI and TH groups who achieved scores at the higher end of the performance spectrum. Additional research that includes (and statistically accounts for) lower-performing young readers is needed, in order to properly capture the relationships between reading and the rhyme effect.

Here, it is worth noting the contrast between the high number of participant exclusions observed in Chapter 5, and the absence of any exclusions reported in a previous study by Coch, Mitra, George and Mitra (2011). Across both studies, children as young as 6 years old took part, and average reading and phonological processing scores were within or above normal limits. Possibly, the discrepancy is attributable to different educational systems in Australia and the US. More specifically, if children in our Chapter 5 study were receiving literacy instruction that strongly emphasised a phonics approach, this may have heightened the ease with which letter-sound knowledge was retrieved. This, in turn, may have made the task of retrieving, storing and comparing the phonological structure of letter names more difficult, and might have interfered with the ERP rhyme effect response for those participants capable of completing the task, as is supported by the negative correlation between letter-sound knowledge and rhyme effect amplitude. Certainly, the relationship between letter-sound knowledge and the rhyme effect is an area worthy of further investigation, as it has not yet been considered in previous studies that have included the letter rhyme judgement task (e.g., Bann & Herdman, 2016; Coch, George & Berger, 2008; Coch et al. 2011).

Despite the CI group's homogeneity in many respects, there was some variability in children's hearing histories, particularly with regard to the age at which they received their implants. Since early cochlear implantation has been associated with better developmental outcomes (e.g., Fulcher, Purcell, Baker & Munro, 2012; Johnson & Goswami, 2010), different results might have been obtained had the criteria for research participation required an earlier age at implantation. It would be beneficial for future studies to evaluate the school-age language and literacy outcomes of children who received their CIs before the age of 12 months. Currently, very little evidence exists for this cohort, and yet it is common practice in Australia for infants as young as 6 months old to receive CIs (Dettman et al., 2016).

Along a similar line of inquiry, given the different hearing experiences of our CI group, it would be worthwhile to directly examine the influence of speech perception on language and literacy results. In particular, nonword repetition and nonword spelling tasks, included as measures in Chapters 2 and 3 respectively, required participants to hear and process unfamiliar phonemic strings. Testing conditions were such that participants were exposed to clear and consistent representations of the target stimuli. Nevertheless, in order to recognise phonological input, it is first necessary for auditory and speech discrimination processes to take place (Stackhouse & Wells, 1997), and it is possible that difficulties here had some bearing on the results. Future research should examine this question by observing the relationship between participants' auditory and speech perception abilities and their performances on nonword processing tasks; indeed, this may be especially important with regard to the implications for repetition. Nonword repetition deficits have previously been thought to represent phonological memory limitations, and thus such difficulties on this task have been attributed to processing failures at a linguistic stage of analysis (e.g., Briscoe, Bishop & Norbury, 2001; Edwards & Anderson, 2014; Nittrouer et al., 2014). Accordingly, additional research is warranted to account for lower-level limitations in the nonword processing performances of children with CIs.

6.7. Overall Conclusions

The aim of this thesis was to provide insight into the literacy development of beginning readers with CIs, who used spoken language only to communicate. As was predicted based on the existing research literature, children with CIs performed poorer than TH children on tasks with high phonological processing demands. Yet, despite the difficulties exhibited by children with CIs, with respect to their spoken and written language skills and sub-skills, the cohort's relative strengths in reading comprehension and spelling accuracy are worth highlighting. Additional longitudinal research would help to address whether these positive outcomes, which are potentially associated with the CI group's high average nonverbal reasoning skills and access to early spoken language-based intervention, are maintained as academic reading and writing demands increase over the school years.

In addition to playing an obvious and fundamental role in engendering academic and vocational success (World Literacy Foundation, 2015), literacy skills are intrinsically tied in with a child's self-efficacy, and their attitude towards learning more generally (Polychroni, Koukoura & Anagnostou, 2006). Hence, where a specific subset of the general population demonstrates a weakness in reading and writing, it is important that continued research efforts are made to examine the potential sources of the deficits, and the extent to which such deficits present in even more narrowly defined subsets of the affected group. While prior studies have established that children with CIs demonstrate literacy difficulties, this thesis extends on existing research by, firstly, examining in further depth the skills that underlie reading and spelling, as defined by existing psycholinguistic-based theoretical models. Secondly, these literacy skills and sub-skills were examined in children with CIs who were homogeneous in age, communication mode, and exposure to behavioural intervention. The findings reported here therefore provide important and unique insights into this cohort's literacy development, on which basis specialised interventions may be developed and evaluated. Ultimately, it is hoped that the research from this thesis contributes to a more complete understanding of what skills can be targeted in the classroom, clinic room and home environments, to best facilitate children with CIs in becoming skilled users of written language.

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Appendices

A. Word-Picture Matching: List of Stimuli (Ch. 4)

CONGRUENT				INCONGRUENT			
1	ant	**31**	key	**1**	axe	**31**	queen
2	balloon	**32**	kite	**2**	bottle	**32**	cave
3	banana	**33**	lamp	**3**	butterfly	**33**	lake
4	bed	**34**	leaf	**4**	boy	**34**	leg
5	belt	**35**	lion	**5**	bar	**35**	lady
6	book	**36**	mouse	**6**	bag	**36**	men
7	boot	**37**	pen	**7**	board	**37**	pants
8	bowl	**38**	pencil	**8**	beach	**38**	puppy
9	bridge	**39**	piano	**9**	bath	**39**	policeman
10	button	**40**	pig	**10**	bedroom	**40**	plum
11	camel	**41**	rabbit	**11**	cabbage	**41**	river
12	candle	**42**	ring	**12**	coffee	**42**	rock
13	car	**43**	ruler	**13**	king	**43**	ribbon
14	carrot	**44**	scissors	**14**	coffin	**44**	sparrow
15	chain	**45**	seal	**15**	chin	**45**	slime
16	chair	**46**	shark	**16**	cheese	**46**	shore
17	clock	**47**	sheep	**17**	cart	**47**	shed
18	cloud	**48**	shirt	**18**	kiss	**48**	sheet
19	cow	**49**	sock	**19**	cake	**49**	skin
20	crab	**50**	spider	**20**	camp	**50**	squirrel
21	crown	**51**	spoon	**21**	coal	**51**	sand
22	duck	**52**	statue	**22**	door	**52**	saddle
23	fish	**53**	sword	**23**	feet	**53**	ski
24	flower	**54**	table	**24**	father	**54**	teacher
25	fork	**55**	tent	**25**	film	**55**	toad
26	fox	**56**	tiger	**26**	food	**56**	trolley
27	hammer	**57**	toilet	**27**	human	**57**	tower
28	hand	**58**	tomato	**28**	hen	**58**	telephone
29	horse	**59**	toaster	**29**	hill	**59**	turtle
30	iron	**60**	tree	**30**	island	**60**	tea

B. Letter Rhyme Judgement: List of Stimuli (Ch. 5)

NO RHYME			RHYME		
1	A	B	**1**	A	J
2	A	C	**2**	A	K
3	A	D	**3**	J	A
4	A	G	**4**	J	K
5	J	P	**5**	K	A
6	J	T	**6**	K	J
7	J	B	**7**	I	Y
8	J	C	**8**	Y	I
9	K	D	**9**	B	C
10	K	G	**10**	C	D
11	K	P	**11**	D	G
12	K	T	**12**	G	P
13	I	B	**13**	P	T
14	I	C	**14**	T	B
15	Y	D	**15**	B	D
16	Y	G	**16**	C	G
17	B	A	**17**	D	P
18	C	A	**18**	G	T
19	D	A	**19**	P	B
20	G	A	**20**	T	C
21	P	J	**21**	B	G
22	T	J	**22**	C	B
23	B	J	**23**	D	C
24	C	J	**24**	G	D
25	D	K	**25**	A	J*
26	G	K	**26**	A	K*
27	P	K	**27**	J	A*
28	T	K	**28**	J	K*
29	B	I	**29**	K	A*
30	C	I	**30**	K	J*
31	D	Y	**31**	I	Y*
32	G	Y	**32**	Y	I*

Note. Letter pairs marked with an asterisk are listed twice, and so were presented four times during the entire experiment (while all others were presented twice). This step was necessary in order for all letters to be presented an equal number of times across prime and target positions, as was a presentation constraint for the experiment.

C. Behavioural Results for Participants with CIs Included in Letter Rhyme Judgement Analyses (Ch. 5)

#	Measure	Score	Significance of difference		Effect size
			t	*p*	(*z*)
1	Real word reading[1]	1.51	0.439	0.666	0.450
	Nonword reading[1]	0.61	-0.123	0.903	-0.126
	Phonological awareness[2]	110	0.022	0.983	0.023
	Phonological memory[1]	0.07	-0.309	0.760	-0.317
	RAN[2]	104	0.171	0.866	0.175
	Letter-sound knowledge[1]	0.04	-0.703	0.490	-0.721
	Nonverbal reasoning[1]	3.22	2.123	*0.047	2.175
	Letter rhyme accuracy[3]	0.85	-0.535	0.599	-0.548
2	Real word reading	-0.20	-0.734	0.472	-0.752
	Nonword reading	0.09	-0.517	0.611	-0.529
	Phonological awareness	100	-0.712	0.485	-0.730
	Phonological memory	0.24	-0.136	0.893	-0.139
	RAN	98	-0.415	0.682	-0.426
	Letter-sound knowledge	-0.10	-0.860	0.400	-0.882
	Nonverbal reasoning	2.42	1.245	0.228	1.275
	Letter rhyme accuracy	0.77	-1.640	0.118	-1.680
3	Real word reading	-0.19	-0.727	0.476	-0.745
	Nonword reading	-0.55	-1.001	0.329	-1.026
	Phonological awareness	92	-1.300	0.209	-1.332
	Phonological memory	-0.89	-1.332	0.199	-1.365
	RAN	95	-0.709	0.487	-0.726
	Letter-sound knowledge	-1.44	-2.363	*0.029	-2.422
	Nonverbal reasoning	1.19	-0.124	0.902	-0.128
	Letter rhyme accuracy	0.84	-0.757	0.458	-0.776
4	Real word reading	-0.92	-1.228	0.235	-1.258
	Nonword reading	-0.77	-1.168	0.257	-1.196
	Phonological awareness	84	-1.888	0.074	-1.934

	Phonological memory	-2.28	-2.798	*0.011	-2.868
	RAN	76	-2.566	*0.019	-2.629
	Letter-sound knowledge	0.36	-0.344	0.734	-0.353
	Nonverbal reasoning	0.01	-1.430	0.169	-1.466
	Letter rhyme accuracy	0.84	-0.757	0.458	-0.776
5	Real word reading	-1.84	-1.859	0.079	-1.905
	Nonword reading	-0.92	-1.281	0.216	-1.313
	Phonological awareness	86	-1.741	0.098	-1.784
	Phonological memory	-0.88	-1.315	0.204	-1.348
	RAN	88	-1.393	0.180	-1.427
	Letter-sound knowledge	-1.25	-2.150	*0.045	-2.203
	Nonverbal reasoning	-0.41	-1.890	0.074	-1.937
	Letter rhyme accuracy	0.89	0.016	0.988	0.016
6	Real word reading	-0.26	-0.775	0.448	-0.794
	Nonword reading	-0.80	-1.190	0.249	-1.220
	Phonological awareness	88	-1.594	0.127	-1.633
	Phonological memory	-1.04	-1.488	0.153	-1.525
	RAN	92	-1.002	0.329	-1.027
	Letter-sound knowledge	-1.25	-2.150	*0.045	-2.203
	Nonverbal reasoning	-0.76	-2.280	*0.034	-2.336
	Letter rhyme accuracy	0.78	-1.528	0.143	-1.566

Note. CI = cochlear implant; RAN = rapid automatised naming. [1]Standardised z-score (normalised test mean = 0.0); [2]Standard Score (normalised test mean = 100.00). [3]Proportion correct across conditions. Statistically significant values (p<.05) indicated with asterisk (*). t- and p-values given to indicate significant difference from control group (n=20) norms; z-value given to indicate effect size, with respect to difference in score from control group norms.